Towards an Explanatory Model of Socio-Emotional Functioning in Children and Adolescents with Congenital Dermatological Disfigurement: The Role of Attachment and Shame

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Abstract

The main aim of this study was to start to develop a framework to understand socio-emotional heterogeneity in young people with congenital dermatological disfigurement. The rationales for this study were a) congenital dermatological disfigurement has been studied far less than other forms of disfigurement b) school-aged young people with disfigurement have been studied less than adults and infants c) the existing literature has indicated a high degree of psychological heterogeneity in this population which has not been adequately accounted for. Specifically, the study investigated the role of attachment and shame in explaining variance in socio-emotional functioning in 8-16 year olds with congenital dermatological disfigurement.

Standardised measures were completed by 122 young people and their accompanying parent/guardian at a specialist paediatric dermatology unit. Comparisons were made between disfigurement group means and general population means. Within participant associations were also explored. The results provided some support for the proposed model.

This study constituted a first step in developing a comprehensive, explanatory framework for understanding socio-emotional development in this population. The study also identified the potential influence of other illness variables and social rejection in explaining socio-emotional functioning in this group.

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1.0

INTRODUCTION

1.1 Section 1: Psychological Functioning and Congenital Disfigurement

The impact of congenital disfigurement on the psychological and social development of a young person is relatively under-researched. Unlike the empirical literature on psychological issues relating to appearance in the non-disfigured majority, which is sizeable and robust, disfigurement research has been sparse and largely focused on specific sub-types of disfigurement. These limitations have affected the usefulness of the existing research for understanding the broader population of young people with disfigurement and in driving the development of effective clinical services. Furthermore, much of the research on the impact of disfigurement has not drawn on the now comprehensive body of research on psychological development, which can provide an empirically supported framework for how a factor such as congenital disfigurement might affect individuals variably.

This study set out to develop the existing literature by addressing some of the knowledge gaps in the field of disfigurement research. Specifically, the aims of this study were to:

1) Investigate psychological functioning in people with congenital dermatological disfigurement, a form of disfigurement which has been under-researched.
2) Investigate psychological functioning in school-aged children and adolescents who have congenital dermatological disfigurement since disfigurement research has focused more on infants and adults.
3) Examine the influence of two psychological factors: attachment and shame on psychological functioning in young people with congenital dermatological disfigurement.

The findings from this study will be useful in facilitating early identification of children who are at risk of developing psychological problems relating to their disfigured appearance, and in facilitating the development of more effective clinical interventions to help those young people who have already developed disfigurement-related psychological difficulties.

1.1.1 Background

The link between physical attractiveness and psychological function is evident in the recorded social narratives of many cultures and across millennia of human civilization.

"The face is the image of the soul"

Cicero, c.106-43 BCE

Modern socio-anthropological and psychological research has sought to develop an understanding of the nature of physical attractiveness and the role of physical attractiveness in society. Substantial empirical support has been found for the hypothesis that physically attractive individuals are viewed and treated more positively by others (e.g., Langlois, Kalakanis, Rubenstein, Larson, Hallam, & Smoot, 2000b) and that, conversely, physically unattractive people are treated less well (e.g., Griffin & Langlois, 2006).

On the basis of the existing literature on the role of physical appearance in social functioning and psychological adaptation, a more recent line of research has emerged that investigates psychological function in people with a disfigured appearance. This field of research is predominantly based on the hypothesis that a disfigured physical appearance affects an individual's social and developmental

experiences negatively and that, therefore, people with disfigured appearance are at increased risk of developing psychological and social difficulties.

The existing body of research on psychological functioning in people with disfigured appearance is still relatively small and largely descriptive. Furthermore, the majority of studies have focused on adults and not children, and on people with acquired disfigurement, such as from burn injuries, rather than people with congenital disfigurement. Moreover, the research on congenital disfigurement has focused predominantly on people with cleft-lip and/or palate (CLP) and, to a lesser extent, on people with congenital cranio-facial anomalies, with very little research on the many other forms of congenital disfigurement. The narrow focus of the existing research limits its usefulness and generalisability. The current study, therefore, aimed to extend the current literature on the psychological impact of physical disfigurement by investigating children, rather than adults, individuals with congenital disfigurement rather than acquired disfigurement and congenital disfigurement resulting from dermatological conditions rather than from CLP. This study further aimed to develop the existing knowledge base by attempting to understand why some individuals, who are congenitally disfigured, appear to fare better psychologically than others (e.g., Rumsey, Clarke, White, Wyn-Williams & Garlick, 2004). To this end, an explanatory model of psychological ontogeny in children with congenital dermatological disfigurement was tested by investigating the association between two key developmental constructs - attachment and shame - and socio-emotional functioning in order to try to explain the psychological variation seen within this population. Overall, the goal of this study was to derive descriptive and explanatory information about psychological functioning in children and adolescents with congenital dermatological disfigurement that could directly inform mental health service provision in order to maximize positive psychological adaptation in this clinical population.

Chapter 1 of this dissertation provides the theoretical and empirical background to the current study as well as outlining the rationale and design of the study.

The first section of Chapter 1:

a) Defines congenital dermatological disfigurement

b) Reviews the existing literature on psychological functioning in children and adults with a disfigured appearance.

The second section of Chapter 1:

a) Introduces the developmental frameworks on which this study is based

b) Introduces the two developmental variables of attachment and shame and explains how they are hypothesised to influence psychological development in children and adolescents with congenital dermatological disfigurement.

The third section of Chapter 1 describes the design of the current study.

1.1.2 Defining disfigurement.

1.1.2.1 Definitional issues.

While the term “disfigurement” is immediately and intuitively understandable in the lay context, there has been much debate about how to operationalise “disfigurement” for the purposes of empirical study in a way that is still intuitively meaningful. Firstly, there has been debate about the use of the term “disfigurement” with critics expressing reluctance to use this term because of a perceived implicit negative value judgement and focus on defectiveness. Researchers have expressed concern that using pejorative terms, such as “mental retardation” which is no longer acceptable as a means of describing people with cognitive function that is significantly lower than population norms, would somehow devalue and undermine those very people who researchers and clinicians are seeking to help (e.g., Rumsey

& Harcourt, 2007b). Proponents of this point of view have argued for the use of the term “visible difference” as a less stigmatizing reference. However, an alternative view, expressed by the Changing Faces charity which supports people with disfigurement (Changing Faces, 2013) is that disfigurement is, inherently, a stigmatized state of being, and to use a more “politically correct” terminology would deny or diminish the lived experience of people who are considered abnormal in appearance by society. It has also been argued that the term “visible difference” diminishes the important qualitative difference between typically acceptable visible differences such as hair texture (e.g., curly or straight) or eye colour (e.g., brown or blue) and stigmatizing visible differences such as burn scarring or significant bony or soft tissue malformation in the case of a disease such as neurofibromatosis. This distinction is referred to by Harris (1997) where he specifies that disfigurement must be a difference that is “non-culturally sanctioned”. It is similarly argued that the term “visible difference” diminishes the quantitative difference between typically acceptable differences such as short and tall height that is within socio-cultural norms compared to a person with achondroplastic dwarfism whose shortness of height is so extreme as to be considered “abnormal” by most socio-cultural standards. For the purposes of the current study, the term “disfigurement” will be used for clarity.

A second issue relating to the definition of disfigurement is the issue of visibility. Many researchers believe that visibility to others is an important feature of disfigurement, because value judgements about a person’s physical appearance can only be made in the context of a social interaction in which the observer makes the judgement that the disfigured person’s appearance is “abnormal” or in which the disfigured person makes the judgement that their own appearance is “abnormal” compared to the appearance of the others around them (Harris, 1997). However, visibility can be a variable state. While a scar on a person’s torso may be invisible

when the person is clothed, it may become visible if the person is lightly clothed, such as in short-sleeves or in a swimming costume, getting undressed in a changing room or during sexual activity. Also, the visibility of several types of disfigurement can vary over the course of time such as in the case of a disease such as eczema or psoriasis. It is unclear whether this population should be considered disfigured all of the time, regardless of the state of their skin condition, only some of the time, when their skin can be clearly observed to be disfigured, or not disfigured at all because, at times, their skin can look normal. There is also evidence to suggest that "hidden" disfigurement can cause a greater sense of shame and distress than visible disfigurement as the individual can fear that their non-visible disfigurement may be inadvertently revealed (e.g., Brown, Moss, Mcgrouther, & Bayat, 2010). Furthermore, much literature on the development of a sense of self, self-worth and self in relation to others is based on the psychological process of an internalised audience (e.g., Lewis, 1971). Therefore, while others may be unaware that a person has a scarred torso, the scarred person will know that they have a scarred torso and they will judge their scarred torso according to the values that they have experienced with the others in their social context.

Harcourt & Rumsey (2008) offered a more concrete and intuitively meaningful definition of disfigurement which removed any value judgement from the meaning; " aesthetic effects of a mark, rash, scar or skin graft on the skin or an asymmetry or paralysis to the face or body". However, without the additional factor of socio-cultural norms, this definition does not differentiate between marks and scars that may be considered normal and non-stigmatizing within a social grouping such as a scar from a tuberculosis vaccine or skin punctures for earrings versus scars or punctures from a traumatic injury or major surgery. Similarly there are many aspects of appearance that are difficult to categorise as a disfigurement based on this definition such as freckles or prominent ears that have been demonstrated to cause

appearance-related distress in the affected individual. It also does not include appearance factors such as short stature that might also mark an individual as looking abnormal.

The definition of the term “disfigurement” to be used for the purposes of the current dissertation will combine the existing definitions as “a difference of physical appearance that is outside the range of what is considered normal within the immediate socio-cultural context, such as aesthetic effects of a mark, rash, scar or skin graft on the skin or an asymmetry or paralysis to the face or body, which is present in either a constant or recurring state”.

1.1.2.2 Congenital and acquired disfigurement.

Disfigurement is often categorized into the quasi-aetiological groupings of acquired disfigurement and congenital disfigurement. Congenital disfigurement has been defined as disfigurement that began “pre-memory” (Harris, 1997), that is, within the first two years of life (Thompson & Kent, 2001). This use of the term "congenital" is at odds with the more common usage of the term meaning present at birth (Medline, n.d.). Acquired disfigurement is any kind of disfigurement that occurs after the first two years of life. Researchers have suggested a difference in psychological functioning in these two groups because the congenital group has never known a life without their disfigurement and the acquired group would have had a period of “normal” psychological development before suddenly having to accommodate the disfigurement into their sense of self and how they fit with the world. Also, acquired disfigurement is sometimes associated with traumatic events such as an accident or major surgery which, it has been argued, might also effect subsequent psychological function (Patrick, Topolski, Edwards, Aspinall, Kapp-Simon, Rumsey, Strauss, & Thomas, 2007). However, the research findings on the psychological functioning of these two groups has not been consistent (e.g., Thompson & Kent,

2001). As such, whether the two groups are psychologically heterogeneous is still unclear.

1.1.2.3 Causes of disfigurement.

Another factor that contributes to definitional problems in disfigurement research is the fact that there are several causes of disfigurement. Disfigurement is a symptom rather than a disease in itself, which means that it has a wide range of manifestations. Thompson & Kent (2001) named three key causes of disfigurement: congenital malformations (e.g., cleft lip and/or palate, port-wine stains), traumatic events (e.g., burns, limb amputations or residual disabilities such as a limp) and disease processes which might be either direct (e.g., dermatological conditions such as acne, eczema or neurological conditions such as stroke resulting in hemiplegia), or indirect (e.g., disfigurement caused by treatment such as surgical scars). Even within Thompson's and Kent's causal groupings, there are significant differences in disfigurement in terms of causal process, course and treatment and therefore it is difficult to determine if an individual affected by one kind of disfigurement, such as a surgical scar on the upper lip, is comparable to someone with another kind of disfigurement such as an amputated leg. Furthermore, many causes of disfigurement can result in other functional difficulties such as with walking, speaking, swallowing or increasing susceptibility to opportunistic illnesses and pain that might also significantly alter the affected individual's life experience and, therefore, their psychological development. There is also diffuseness in categorisation. For example, while congenital malformations, by definition, must also be congenital disfigurements, disfigurements related to traumatic events and disease processes could be either congenital disfigurements or acquired disfigurements depending on whether they occur during the "pre-memory" phase of life as per the definition of congenital disfigurement (Thompson & Kent, 2001) or later in life.

1.1.2.4 Epidemiology.

Estimating the prevalence and incidence of disfigurement is very difficult, primarily due to the definitional issues described earlier. Estimates have varied significantly. Based on UK census information, it was estimated that approximately 1% or 600,000 people were disfigured (Office of Population Census & Surveys, 1988, cited in Thompson & Kent, 2001). In an unpublished study, Changing Faces estimated that 1,345,000 people in the UK, or 1 in 44, are disfigured (Julian & Partridge, 2007) while Rumsey (1998) estimated an even higher figure of 10% of the UK population which is approximately six million people, referencing prevalence studies on medical conditions that are disfiguring rather than including only studies of individuals who identify themselves as disfigured. For example, in a recent community-based study, Smithard, Glazebrook & Williams (2001) found that 50% of UK adolescents were diagnosed with mild to severe acne, a significant proportion of whom reported psychological difficulties relating to their appearance.

1.1.3 Disfiguring dermatological conditions.

Dermatological conditions are inherently disfiguring. The skin is the largest and most observable organ of the human body. Skin has the potential to express important information about the general health of the individual. Researchers have argued that there are several aspects of the skin that convey general health, ageing and fertility of the individual which have become translated into indices of physical attractiveness (e.g. Samson, Fink & Matts, 2010; Furnham, Mistry, & Mcclelland, 2004).

There is a wide range of dermatological conditions seen in children and young people. The most common of these, such as Atopic Eczema (AE), are some of the most common diseases seen in childhood. Eczema has a lifetime prevalence at age

10 years of 41% in UK children (Nice, 2007). Two percent of these cases are rated as severe by a dermatologist, with the vast majority being rated as mild (NICE, 2007) Birthmarks are also a very common dermatological condition with port-wine stains, just one type of lesion commonly referred to as a birthmark, affecting 3-5 per 1000 live births (Alper & Holmes, 1983). Other dermatological conditions such as Epidermolysis Bullosa (EB) (1/50,000 live births) and Overgrowth Syndromes such as Neurofibromatosis Type 1 (NF1) (1/2500-3300 live births) are much more rare and are largely managed in highly specialist dermatological services (Schofield, Grindlay, & Williams, 2009). The onset of dermatological disease is variable. Genetic and congenital conditions, such as EB, NF1 and birthmarks, tend to be diagnosable at birth (although sometimes diagnosis occurs slightly later due to disease development and manifestation). Other common dermatological conditions such as acne, warts, psoriasis, vitiligo, alopecia areata and eczema can occur at various times across the lifespan.

Dermatological conditions, while inherently disfiguring due to their manifestation on the skin, are complicated by a range of other concomitant symptoms. Most commonly there is discomfort relating to pain or itching. Some conditions are also associated with significant mobility difficulties such as NF1 or are potentially life limiting such as EB.

There is a wide range of severity and treatability across dermatological conditions. Many dermatological conditions do not have a medical cure. As such, many are present in some form throughout the lifespan. Some conditions spontaneously remit over time, such as in the case of some childhood eczema. However, many of the more complex dermatological conditions have symptoms that can be minimized but will otherwise be present constantly or recurrently throughout life, such as psoriasis or EB.

1.1.4 Psychological functioning in people with disfigurement.

1.1.4.1 Adults with disfigurement.

The majority of large-scale studies on psychological functioning and disfigurement have been conducted with adults with both acquired and congenital disfigurement. Studies with children and adolescents are relatively few in number. Several studies of disfigured adults have reported no major psychological maladjustment (e.g., Van Den Elzen, Versnel, Hovius, Passchier, Duivenvoorden, & Mathijssen, 2012) . However, some studies have reported evidence of impaired quality of life and difficulties in a range of areas of functioning. The most commonly identified areas of difficulty have been negative self-perceptions, emotional problems, and social difficulties (e.g., Hunt, Burden, Hepper, & Johnston, 2005; Stubbs, James, Daugherty, Epperson, Barajaz, Blakeney, Meyer Iii, Palmieri, & Kagan, 2011). The findings of the largest, more recent studies with disfigured adults are described below.

Rumsey, Clarke, White, Wyn-Williams & Garlick (2004) conducted a large-scale study of 458 adults drawn from 15 different hospital clinics dealing with disfiguring conditions. The study used one normed psychological measure of depression and anxiety: the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983). The results indicated that the overall sample mean was below clinical cut-off for both anxiety and depression. However, 48% of the sample reported anxiety levels in the borderline to abnormal ranges and 27.5% reached borderline or caseness levels for depression. These percentage scores were both elevated compared to the normal population estimates of 38% and 23% respectively (Crawford, Henry, Crombie, & Taylor, 2001) although the significance of these differences in scores was not statistically tested. The authors also reported a wide range of scores across study sub-samples. The study further evaluated the

relationship between disfigurement severity and psychological functioning. There was only one significant difference by disfigurement severity. The group with the least noticeable disfigurement reported significantly higher levels of anxiety than the three more severe groups. There were no other significant differences according to severity grouping such as on measures of quality of life, depression or social avoidance and social distress.

van den Elzen et al. (2012) conducted a study on social functioning in 118 adults with facial disfigurements. Fifty-nine had congenital disfigurements related to CLP and 59 had acquired disfigurement as a result of a traumatic accident. All disfigured participants were sampled from two large specialist hospitals in the Netherlands. A non-disfigured comparison group was sampled from a number of general medical practice clinics in the same city as well as from a university student population. The authors aimed to evaluate the impact of acquired and congenital disfigurement on social functioning. The Social Avoidance and Distress Scale (Watson & Friend, 1969) and the Scale for Interpersonal Behaviour (Arrindell, De Groot, & Walburg, 1984) were the two psychological measures used.

The study found that there were no significant differences between the congenital and acquired groups on any whole psychological measures. The combined disfigured group did not report significantly different levels of social anxiety and distress compared to the comparison group. However, the congenitally disfigured group scored significantly lower than the other two groups on three subscales of the Scale for Interpersonal Behaviour: expressing negative feelings, initiating assertive behaviour and expressing personal limitations. Both disfigurement groups also reported lower satisfaction with appearance than the non-disfigured group. Satisfaction with appearance was significantly associated with all social functioning subscales except for frequency of expressing positive feelings. Objectively

assessed disfigurement severity was not associated with any psychological measures.

The van den Elzen et al. (2012) study was interesting in reporting that, while the disfigured participants were no more likely to reach diagnostic levels of social difficulty as measured by the Social Avoidance and Distress Scale, there were indications of more subtle difficulties in social functioning. This study did not further investigate the range of response in the same way as the Rumsey et al (2004) study which also found their disfigurement group means to be unremarkable but then discovered a higher rate of caseness in their disfigured sample. van den Elzen et al. (2012) expressed some concern about the validity of their comparison sample, however, questioning whether the inclusion of so many students from a prestigious university might have skewed the psychological scores of their non-disfigured group. Certainly, the non-disfigured group had a much higher level of education than the two disfigurement groups.

Both studies demonstrated the complexity of the psychosocial picture in adults with disfigurement. There is some evidence to suggest an increase in psychological problems in the disfigured population. However, the findings were equivocal. There were differences in rates of reported psychological difficulty across some aetiological groupings but not others. Differences between the disfigured population and the non-disfigured population were evident on some psychological indices, but not others. Despite the large total sample sizes, both studies had heterogeneous samples that may have introduced error variance into the results as indicated by some significant within-group differences.

1.1.5 Systematic review of studies of psychological functioning in children and adolescents with congenital disfigurement.

In a recent position paper, Rumsey & Harcourt (2007a) reported that the impact of having a disfigured appearance during childhood and adolescence was still equivocal. The authors reported that studies had found contradictory results about the psychological functioning of disfigured young people, with some papers reporting significantly poorer function compared to the non-disfigured population (e.g., Absolon et al., 1997) while others had reported no difference, and in some papers, significantly better functioning compared to the normal population (e.g., Walters, 1997) .

Currently, there are no published systematic reviews of the existing literature on children and adolescents with disfigurement. Many key publications have been predominantly position or editorial pieces or conceptual treatises focusing on the phenomenology of disfigurement and expert opinion on intervention (e.g., Newell, 2000; Rumsey & Harcourt, 2007b). It has been more difficult to identify a robust body of peer-reviewed empirical papers on young people with disfigurement. As such, it was considered that an important first step in the current study was to systematically review the most recent literature on the psychological impact of congenital disfigurement in children and adolescents.

The current review focused on congenital disfigurement in order to reduce the heterogeneity of the population. Previous researchers have argued that the psychological adjustment process to acquired disfigurement might be significantly different to that relating to disfigurement that has been present for as long as the individual can remember (Thompson & Kent, 2001). The review also focused on 8-16 year olds as this is the age range that is most commonly seen in child and adolescent mental health services. Developmentally, this age range could also be

considered the period during which children become increasingly aware of themselves in relation to their peers and the wider social context and whether they are considered to be socially valuable by their peers (Erikson, 1959). The search also included only quantitative studies in order to maximize the comparability and robustness of findings across papers.

1.1.5.1 Method.

The question for review was “What is the psychological and social functioning of 8-16 year olds with congenital disfigurement?”

The full search protocol is contained in Appendix A. The PICOS model (CRD, 2009) was used to form the basis of the current search protocol. In brief, five electronic databases were included in the search; Pubmed, PsycINFO, PsycEXTRA, CINAHL and Web of Science (See Appendix B for Search Strategy). The papers fitting the search criteria were identified as shown in Figure 1. The key information from the included papers was summarized using a Data Extraction Form (Appendix C) and entered into Table 1.1 and into the summary table of additional information (Appendix D). The quality of the papers was evaluated on the basis of the following criteria which were based on the criteria recommended by the Cochrane Handbook (Reeves, Deeks, Higgins, & Wells, 2008) for evaluating quality in studies that are not randomised controlled trials of interventions:

1) Adequate Sample Size.

2) Representative Sampling Method.

3) Validity and reliability of outcome measurement.

4) Validity of comparison group.

5) Appropriateness of statistical analysis.

6) Reporting biases.

7) Any other biases.

Details of the quality assessment protocol are described in the search protocol in Appendix A.

1.1.5.2 Results

1.1.5.2.1 Search strategy results

The initial search of the electronic databases yielded 2450 references. Using the duplicate function on EndNote®, 1602 duplicates were removed. Four hundred and ninety six irrelevant papers were removed and a further 342 were removed because they did not fit the search criteria. Ten papers remained in the review. A hand search was then conducted of references and citations from each of these 10 papers which elicited a further two studies which fitted the review criteria. Twelve papers were included in the final review. Figure 1.1 summarises the process of attrition of search results. Table 1.1 summarizes the studies included in the review. A glossary of acronyms used in Table 1.1 is shown in Table 1.2.

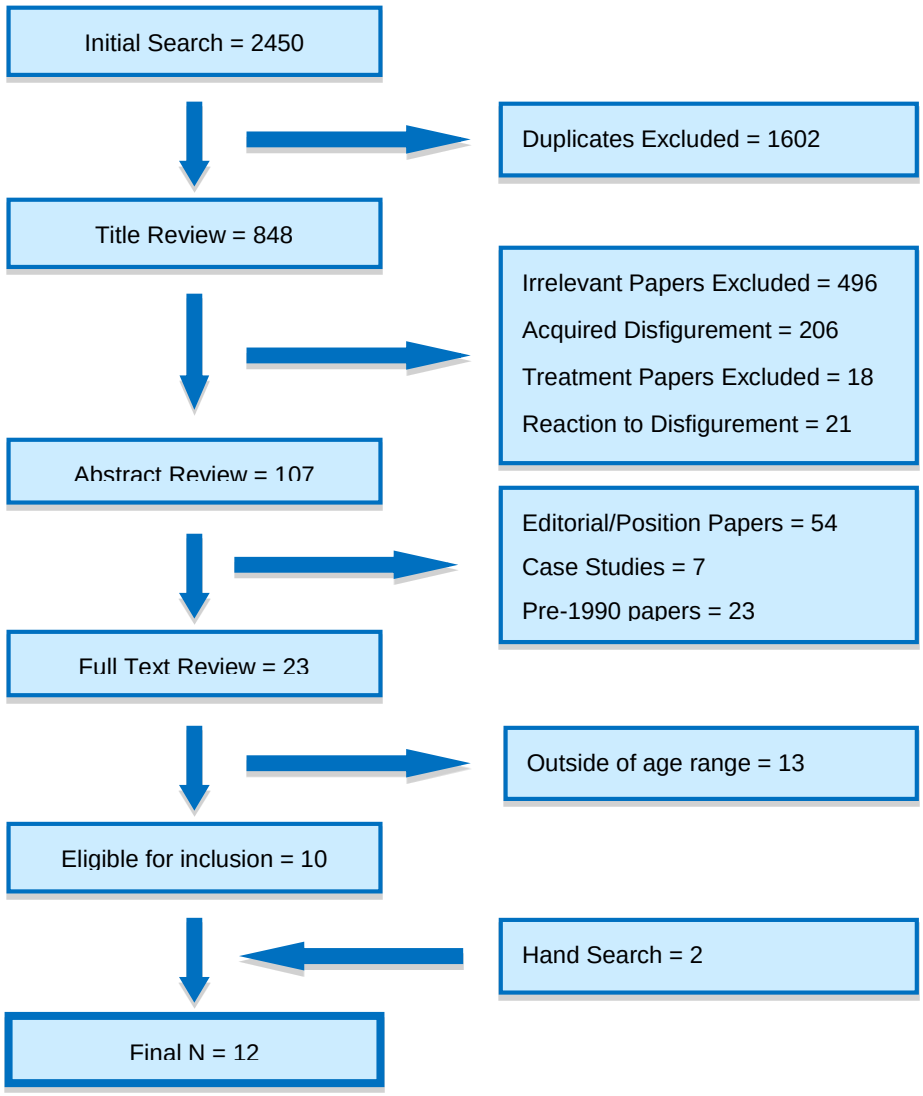

Figure 1.1 Flow Diagram of Search Strategy Results

Table 1.1

Summary of Papers (See Table 1.2 below for Glossary of acronymns)

Legend for Comparison Findings
Disfigurement Group scored significantly worse than comparison group
Disfigurement Group scored significantly better than comparison group
NS: No significant difference between disfigurement and comparison groups

Identifiers		Sample			Design			Results
Authors	**Year**	**Age Range**	**Sample Size**	**Type of disfigurement**	**Measures**	**Reported by**	**Comparison Group**	**Group Comparison Findings (See Appendix C for other key findings)**
Andersson, Gillberg, Fernell, Johansson & Nachemson	2011	9-11 (M=10.6)	92	CHD	PH	child	school (n=49)	**PH:** Mild disfigured group significantly worse on popularity Severe disfigured group significantly better on intellectual and school status
Feragen, Kvalem, Rumsey & Borge	2010	16	289	CLP	HSCL-25 CF SAc PA	child	Epidemiological (General community) (n=1832)	**SPP:** Visible cleft group on CF, SAc. CLP on PA **HSCL-25:** visible cleft group on emotional distress
Shute, McCarthy, Roberts	2007	11-14 (M=12.6)	48	CFS, CLP, CPO	SPP SASA CBCL	child parent	Published norms	**SPP:** NS **SASA:** NS **CBCL:** internalizing problems, social problems
Topolski, Edwards, Patrick	2005	11-18 (M= not specified)	56	CLP, CFA, AD	CDI YQOL-R	child	C1: community (n=116) C2: mobility limitations (n=52) C3: ADHD (n=68)	**CDI:** NS **YQOL-R:** Overall QOL, peer interactions, environmental/cultural QOL
Millard & Richman	2001	8-17 (M=11.7)	65	CLP-U, CLP-B, CPO	RCMAS, RCDS/RADS, PBS-D/A, PBS-C, PBS-Cog,	child parent, teacher	Published norms	**RCMAS:** not compared **RCDS/RADS:** not compared **PBS-D/A:** CLP-U, CLP-B, CPO **PBS-C:** CLP-U, CLP-B, CPO **PBS-Cog:** CLP-U, CLP-B, CPO

Identifiers		Sample			Design			Results
Authors	**Year**	**Age Range**	**Sample Size**	**Type of disfigurement**	**Measures**	**Reported by**	**Comparison Group**	**Group Comparison Findings (See Appendix C for other key findings)**
Pope & Ward	1997	11- 13 (M=12.7)	24	CFS, CPO, CLP	SPP CBCL	child parent	Published norms	**SPP:** NS **CBCL:** NS
Richman & Millard	1997	12	44	CLP, CPO	BPC	parent	School (n=667)	**BPC:** girls on internalizing, conduct disorders
Sheerin, MacLeod & Kusumakar	1995	7-15 (M=11.0)	79	PWS, PE	SPP, RCMAS, CDI CBCL	child parent	Matched school group (n=80) Published norms	**SPP:** PWS on SchA, SAc, GSW PE on SchA, AC, PA **CBCL:** PE on social problems, attention **RCMAS:** NS **CDI:** NS
Campis, DeMaso & Twente	1995	6-12 (M=8.1)	77	CLP, CFA, VA	CBCL	parent	Published norms	**CBCL:** NS
Kapp-Simon, Simon & Kristovich	1992	10-16 (M=12.3)	45	CLP, CFA	SPP PIC-A, PIC-SS, RBPC-AW	child parent	Published Norms	**SPP:** NS **PIC-A:** CFA significantly worse **PIC-SS:** NS **RBPC-AW:** NS
Leonard, Brust, Abrahams & Sielaff	1991	8-18 (M=12.0)	105	CLP, CLO, CPO	PH	child	Published norms	**PH:** Mean popularity score below norm
Padwa, Evans & Pilleme	1991	6-16 (M=not specified)	30	CFA, CFS	CDI, HFD, TED CBCL	child teacher	Published norms	**CDI:** NS **HFD:** not compared **TED:** not compared **CBCL:** NS

Table 1.2
Glossary of Acronyms

Type of Disfigurement		Questionnaires Used	
AD	acquired disfigurement	BPC	Behaviour Problem Checklist
CFA	craniofacial anomaly	CDI	Child Depression Inventory
CHD	congenital hand deformities	CFSEI	Culture-free self-esteem inventory
CFS	craniofacial syndrome	CMBSS	Chinese Miller Behavioural Style Scale
CLP	cleft lip/palate	CogAT	Cognitive Abilities Test
CLP-B	bilateral cleft lip/palate	HFD	Human Figure Drawing
CLP-U	unilateral cleft lip/palate	HSCL-25	Hopkins Symptom Checklist – 25
CPO	cleft palate only	PBS	Pediatric Behaviour Scale
PE	prominent ears	PH	Piers-Harris Self-Concept Scale for Children
PWS	port-wine stain	PIC	Personality Inventory for Children
VA	vascular anomaly	RADS	Reynolds Adolescent Depression Scale
		RBPC	Revised Problem Behaviour Checklist
		RCDS	Reynolds Child Depression Scale
		RCMAS	Revised Child's Manifest Anxiety Scale
		SADS	Social Avoidance and Distress Scale
		SPP	Harter Self-Perception Profile
		AC	SPP-Athletic Competence Subscale
		CF	SPP-Close Friendships Subscale
		GSW	SPP-Global Self-Worth Subscale
		PA	SPP-Physical Appearance Subscale
		SchA	SPP-Scholastic Achievement Subscale
		SAc	SPP-Social Acceptance Subscale
		SWLS	Satisfaction with Life Scale
		TED	Tasks of Emotional Development
		YQOL-R	Youth Quality of Life Instrument – Research

1.1.5.2.2 Summary of papers.

Twelve papers published between 1990 and 2013 were included in the review. Ten of these papers studied the cleft lip and/or palate (CLP) population and four out of these 10 included cranio-facial anomalies (CFA) also. Only two papers investigated other congenitally disfigured populations; port-wine stains (PWS) and prominent ears (PE) in one study (Sheerin, Macleod, & Kusumakar, 1995) and chronic hand deformity (CHD) in the other (Andersson, Gillberg, Fernell, Johansson, & Nachemson, 2011).

All studies were questionnaire-based. The study by Padwa, Evans & Pillemer (1991) also included non-validated projective tests. Nineteen different outcome questionnaires were used across the 12 papers with the Child Behaviour Checklist (CBCL; Achenbach & Edelbrock, 1984) and Harter Self-Perception Profiles (SPP; (Harter, 1985; 1988) being the most commonly used questionnaires, each being used by five different studies.

Four papers used parent and child reported outcomes (Shute et al., 2007; Pope & Ward, 1997; Sheerin et al., 1995; Kapp-Simon et al., 1992), four used child-reported outcomes only (Andersson et al., 2011; Feragen et al., 2010; Topolski, Edwards & Patrick, 2005; Leonard et al., 1991) and two papers used parent-reported outcomes only (Richman & Millard, 1997; Campis, DeMaso & Twente, 1995). Millard & Richman (2001) used parent, child and teacher reported outcomes and Padwa et al (1991) used child and teacher reported outcomes.

Eight of the studies were conducted in the US with one study each conducted in the UK, Australia, Sweden, Norway and Hong Kong. Two studies included only "primary school-aged" children, who were younger than 11 years old. Seven studies included only "adolescents". While the age ranges defined as adolescence varied between

studies, in combination, they covered the ages of 10 to 18 years. Four studies combined both primary and secondary school-aged participants.

All papers reported comparative data. Ten studies investigated within disfigurement-group differences as well as differences between the disfigurement group and a comparison group. These within group variables included gender and age, diagnostic categories or psychological variables (See Appendix D for a tabular summary of within-group data).

A quantitative analysis of the findings of the papers included in the literature review was not conducted for several reasons. First, the papers included in the review investigated a range of different domains of psychological functioning such as self-concept, internalizing disorders, quality of life, conduct, school and intellectual attainment and popularity. Second, a number of different measures were used to investigate the differences between the disfigurement sample and the general population. Third, a wide range of findings was reported with the disfigurement groups reporting significantly better and worse functioning than the general population and, in most cases, no significant differences being found. Fourth, some of the older papers did not report sufficient information to calculate effect sizes from their reported results. Fifth, many of the significant results reported were of sub-groups of the disfigurement sample while whole group findings were not significant. Calculable effect sizes varied greatly. For example, Andersson et al (2011) reported small-medium effect sizes such as on popularity, d=0.47, with the disfigured group reporting worse popularity than the general population group. Medium to large effects were reported on general quality of life η^2=.13 by Topolski et al. (2004) with the facially disfigured group reporting significantly worse general quality of life than the non-disfigured group. A large effect size on a comparison of social difficulties, d =1.01445, was reported by Sheerin et al. (1995) with the group with prominent ears

reporting worse social functioning than the non-disfigured comparison group and on global self worth *d* = 1.01445 (Sheerin et al., 1995) with the group with port-wine stains reporting significantly better self-worth than the comparison group.

1.1.5.2.3 Quality assessment.

The papers included in this review demonstrated positive and negative quality attributes.

1) Sample Size: In relation to the statistical analyses used, most studies had adequate to large sample sizes for their group comparison analyses, although some studies had notably uneven group sizes, which may have affected the reliability of the parametric, between group analyses that formed the basis of most of the reported studies. Padwa et al (1991), Pope & Ward (1997) and Millard & Richman (2001) had between 20 and 30 participants per disfigurement group (Millard and Richman divided their total disfigured sample of 65 into three groups based on disfigurement type). These were the smallest samples of the studies included in this review. Given that few studies on disfigured children and psychological functioning has found large effect sizes when comparing disfigured with non-disfigured groups, it is unlikely that these sample sizes would be sufficient to pick up on meaningful between group differences. None of these studies provided information about whether their sample sizes were expected to provide adequate power. No data were provided on the representativeness of these small samples which might also affect the reliability and validity of any findings reported.

2) Sampling Procedure: All studies used samples drawn from highly specialist medical clinics specifically established and funded to meet the needs of patients with congenital anomalies. It was not possible to determine from the papers if there were children with the same disfiguring conditions who were

managed in non-specialist services who were not included in the studies. Not all studies reported the proportion of their sample populations that participated in the study. As such, for many studies, it was impossible to determine if the reported sample was representative of the population within the specialist clinics being sampled. In those studies that reported the proportion of inclusion, one study reported a very low proportion of inclusion of 17% (Shute, Mccarthy, & Roberts, 2007) whilst the remainder reported a range from 51% to 100% participation. Of the 12 studies included in this review, only one study (Sheerin et al., 1995) was conducted in the UK. Seven of the studies were conducted in the US and the remainder in Australia, Sweden, Norway and Hong Kong. As such, the generalisability of the findings from all but one of the included studies may be limited when attempting to understand the UK population better.

3) Reliability and Validity of Outcome Measures: All studies used standardised measures with reported adequate reliability and validity for between group comparisons. Only two studies utilized non-standardised measures (Padwa et al., 1991; (Richman & Millard, 1997) but this was for descriptive purposes only.

4) Validity of Comparison Group: Five of the 12 studies used school or community-based comparison groups. Only one study constructed a matched control group, based on demographic variables. The remainder utilized published norms only. No patterns of results emerged that might have been attributable to the nature of the comparison group.

5) Use of Appropriate Statistical Analyses: All but three studies used appropriate statistical analyses to test group comparisons. In the studies of

Richman & Millard (1997) and Millard & Richman, (2001) no form of statistical analysis was used to compare disfigurement group means with the non-disfigurement comparison group data despite the authors interpreting between group differences in their results.

Shute et al (2007) used appropriate group comparison analyses but also reported increased risk data without using a categorical statistical analysis to determine if the increased risk was significant.

6) Reporting Bias: There was a tendency to report and focus on findings of significant differences, even when findings were mixed or where significant findings were only found on a limited number of individual subscales. Few papers chose to focus on the fact that their findings were mixed or, in some instances, that their findings indicated better adjustment in the disfigured population.

7) Other Biases: All but two studies used CLP and CFA samples limiting generalisability to other forms of disfigurement. All but one study (Millard & Richman, 2001) excluded people with learning disability as a possible confounding variable and all studies controlled the heterogeneity of their sample by limiting their participants to between one and three medical conditions that were the cause of the disfigurement and limiting the age range of their sample. The Millard & Richman (2001) study was the only study to include participants with mild learning difficulties, which may have skewed their findings. Few papers reported on co-morbid symptoms experienced by participants. One key failing was that the majority of the studies were not hypothesis driven. Rather, they were exploratory studies

that used a large number of outcome measures. As a result, the findings were mixed and diffuse and difficult to draw conclusions from.

1.1.5.2.4 Findings: Comparison between the disfigured and non-disfigured groups. Seven of the studies relied on published norms to represent the non-disfigured population (Campis, Demaso, & Twente, 1995; Kapp-Simon, Simon, & Kristovich, 1992; Leonard, Brust, Abrahams, & Sielaff, 1991; Millard & Richman, 2001; Padwa et al., 1991; Pope & Ward, 1997; Shute et al., 2007). The remaining five studies (Andersson et al., 2011; Edwards, Patrick, Topolski, Aspinall, Mouradian, & Speltz, 2005; Feragen & Borge, 2010; Richman & Millard, 1997; Sheerin et al., 1995) constructed their own comparison groups either from existing data sets or by collecting comparison group data for the specific purpose of the study.

Three studies found no significant differences between group means on any outcome measures (Pope & Ward, 1997; Campis et al, 1995; Padwa et al, 1991). These studies used the self-reported CDI and SPP measures and the parent reported CBCL. The Pope and Ward (1997) and Padwa et al (1991) studies are notable in that their sample sizes were the smallest of the studies included in this review. At 24 and 30, respectively, these samples were small for even simple between group comparison analyses. As such, these studies may not have been adequately powered to pick up between group differences. Furthermore, each sample included two or three types of disfigurement. This within group heterogeneity may have further contributed to difficulties in statistically identifying between group differences. The Padwa study reported significant within group differences, which adds further weight to the hypothesis that within group heterogeneity could have obscured between group differences. Padwa et al (1991) also used unvalidated projective tests in their studies from which the authors concluded that their disfigured sample showed a consistent pattern of low self-esteem and denial,

particularly of social problems. The authors hypothesized that these psychological processes may have been associated with underreporting of symptoms on the questionnaire measures, which might have accounted for the lack of significant difference between their disfigured and non-disfigured groups.

Four studies found that all, or specific subgroups, of their disfigured sample scored significantly better than the normal population on some psychological outcomes (Andersson et al., 2012; Feragen et al., 2012; Millard & Richman, 2001; Sheerin et al, 1999). The Millard & Richman (2001) study was unique in that it was the only study to find significantly better results for the disfigured group on parent-reported measures. However, this was the only study in this group which did not use statistical analysis to interpret differences between disfigurement group scores and published norms. As such, their conclusions about differences between disfigured and non-disfigured populations should be treated cautiously. Furthermore, while their total sample size of 65 is sufficient for a single sample, the authors divided their 65 participants into three groups defined by type of disfigurement; unilateral cleft lip, bilateral cleft lip and cleft palate only. All comparisons were made using these three groups meaning that the size of each group was relatively small for statistical analysis.

Sheerin et al (1994) found that the port-wine stain (PWS) group reported significantly better scores than the norm on the self-reported scholastic achievement, social acceptance, global self-worth subscales of the SPP, although the prominent ears (PE) group scored significantly lower than the norm on scholastic achievement, athletic competence and physical appearance. Feragen et al (2012) found that the disfigurement group self-reported significantly better close friendships, social acceptance and satisfaction with physical appearance than the non-disfigured comparison group. There were no other significant differences

between the disfigured and non-disfigured groups in the Feragen study. Andersson et al (2011) reported that their severely disfigured group reported themselves to have significantly better school and intellectual status than the study's non-disfigured group although the mildly disfigured group reported significantly lower popularity than the non-disfigured comparison group.

The Feragen team attributed their finding of more positive psychological functioning in their disfigured group to increased psychological resilience due to being exposed to the stress and challenge relating to their disfigurement during the developmental period. The authors argued that being forced to live with their disfigurement meant that the affected children developed better coping strategies than their non-disfigured peers. Similarly, the Andersson research group attributed their findings of better psychological functioning in the most severely disfigured group compared to the mildly disfigured group to the severe group being forced to acknowledge and cope with the reality of their disfigurement thereby coming to some degree of resolution. In contrast, mildly affected individuals, who could sometimes hide their disfigurement or could perceive themselves to be "nearly normal" were either living in constant fear of being discovered to be different or had never learnt how to constructively accommodate their disfigurement into their sense of self and place in the world. The idea of young people becoming psychologically more resilient as a result of being exposed to stress in early life is well-known, such as in the case of the developing literature on post-traumatic growth (e.g., Hefferon, Grealy, & Mutrie, 2009). However, all studies, except the Millard and Richman (2001) study which did not statistically analyse their findings, in which the disfigured group reported significantly better scores than the normal comparison group used child self-reported outcomes. As such, it is unclear whether there is a response bias in self-reported outcomes or if these findings are, indeed, indicating better than normal psychological adjustment in young disfigured people. It would have been interesting

to compare those child reported outcomes against outcomes reported by others, especially since there is a consistent bias in the disfigurement and general paediatric literature for the child patients to report better functioning than the norm and for their parents, at the same time, to report poorer functioning in their sick children compared to the norm (e.g., Phipps & Srivastava, 1997).

Eight of the 12 studies reported that the whole disfigurement group, or subsets of the whole disfigurement group, reported significantly worse outcomes than the non-disfigured comparison group (Andersson et al., 2011; Shute et al., 2007; Sheerin et al., 1995; Leonard et al., 1991; Topolski et al., 2005; Kapp-Simon et al.,1992; Richman & Millard, 1997; Millard & Richman, 2001;). Andersson et al (2011) reported that the mildly disfigured group reported significantly worse popularity than the moderately disfigured, severely disfigured and non-disfigured groups in the study. Sheerin et al (1995) found that the PE group scored significantly worse on scholastic achievement, athletic competence and satisfaction with appearance on the SPP compared to the matched control group and on social problems and attention problems on the CBCL compared to published norms. Leonard et al (1991) reported that, while their disfigured group did not differ significantly on the overall score of the PH, they scored significantly worse than the published norms on the popularity subscale. Topolski et al (2005) reported that their disfigurement group scored significantly worse than the school-based comparison group on quality of life, particularly in the domains of peer interactions and environmental and cultural quality of life which the authors attributed to a sense of lack of safety at school and in public. While the study conducted by Shute and colleagues (2007) found no significant differences based on group means, they reported that an increased rate of disfigured participants scored in the clinical ranges on the internalizing problems scale (42%) and the social problems sub-scale (33%) compared to the normal population. The authors concluded that there was a substantially higher proportion

of individuals in this population that were suffering from psychological difficulties but, due to the wide variation in functioning within the group, the high number of participants scoring in the clinical range was hidden when only looking at group means. This increased risk of caseness reported in the disfigurement group was not statistically analysed and it is, therefore, not clear if this is a meaningful difference. Kapp-Simon et al (1992) compared their disfigurement group means with published t-scores on the self-reported SPP, the parent-reported Adjustment and Social Skills subscales of the PIC and the parent-reported Anxiety-Withdrawal scale of the RBPC. They reported that the disfigurement group scored 1.5 standard deviations above the published mean on the PIC adjustment scale, which the authors interpreted as a significant difference. They calculated that 59% of their sample were within the clinical range for that measure. There were no significant differences on the self-reported SPP scales or the parent reported social skills and anxiety-withdrawal measures. Richman & Millard (1997) reported that girls in the disfigurement group scored significantly worse than the school group on the internalizing disorders and conduct disorders scales of the parent-reported BPC but that there were no differences between boys in the two groups. The same authors later reported (Millard & Richman, 2001) that the CPO group scored significantly higher on anxiety (RCMAS) and depression (RCDS-R) than the published norms. However, the other two CLP groups scored significantly lower on the same measures compared to the published norms. These two studies conducted by Richman and Miller reported between group differences that were not supported by statistical analyses as well as being based on small sub-group sample sizes. As such, their conclusions about group differences should be considered with caution.

Conclusions

Overall, in the studies reviewed, significant difficulties fell mainly into the domains of social difficulties, quality of life and internalizing difficulties, and were found on both

parent and child reported measures. However, all significant differences were reported alongside a greater number of outcomes that were not significantly different between disfigured and non-disfigured groups. Importantly, there was marked psychological heterogeneity within the disfigured population, with significant differences reported in subgroups of the disfigurement sample and a number of psychological outcomes on which some disfigurement samples scored better than the comparison group.

1.1.5.2.5 Additional data.

As well as conducting means comparisons between disfigured group(s) and non-disfigured comparison groups, many studies reported other data, particularly within-disfigurement group comparisons. While not directly answering the objective of the review, the results from the other analyses are reported below as they provide relevant information regarding disfigurement and psychological functioning (See Appendix C for Tabular presentation of the data described below).

Disfigurement Severity

One study, (Andersson et al., 2012), reported that the mild disfigurement group obtained a significantly worse score than the severe disfigurement group on the overall score of the Piers-Harris Self-Concept Scale for Children, as well as in the sub-domains of behaviour, school and intellectual functioning and popularity. This study was unique in the review because it studied hand deformities rather than disfigurement of the face and head. The nature of the disfigurement and the way in which severity of the disfigurement was measured were different to the remaining studies that focused on populations with face and head disfigurement, which may account for the findings. Four studies compared their samples on severity of disfigurement (Shute et al., 2007; Sheerin et al., 1995; Campis et al., 1995;

Andersson et al., 2011). Three of the four studies found no associations between disfigurement severity and psychological outcome.

Medical Condition

Three papers compared their sample based on the medical condition underlying their disfigurement. Kapp-Simon et al (1992) compared CFA and CLP participants and found no significant differences between the two groups on any psychological outcome measures. Millard & Richman (2001) compared participants with cleft palate only (CPO) with a unilateral CLP group and a bilateral CLP group. They reported that the CPO group scored significantly worse on measures of anxiety and depression. However, this study also determined that the CPO group was more likely to report mild or specific learning difficulties (patients with significant developmental delay had been excluded from the study), which might account for poorer psychological functioning. The study also reported that psychological outcome in the CPO group was more strongly associated with speech functioning than in the other two disfigurement groups. However, there were only approximately 20-25 participants in each of these three disfigurement groups. Furthermore, between group differences were not statistically analysed. Therefore, the study findings were difficult to interpret.

Sheerin et al (1995) compared their port-wine stain (PWS) group against their prominent ears (PE) group and found that the PE group reported significantly poorer scores than the PWS group on scales of self-perception, concentration anxiety, social withdrawal, social problems and internalizing and externalizing symptoms. This finding was interesting as prominent ears, on a prima facie level, could be argued to be generally considered to be less "pathological" than a facial port-wine stain.

Nature of the Disfigurement

Two studies conducted within group comparisons based on one particular feature of the disfigurement. Padwa et al (1991) compared symmetrical against asymmetrical craniofacial anomalies. The study found that the group with symmetrical disfigurement scored significantly more poorly on the Child Depression Inventory and Child Behaviour Checklist, contrary to their initial hypothesis. Feragen et al (2010) split their CLP group into visible disfigurement and non-visible disfigurement subgroups. The visible disfigurement group scored significantly better than the non-visible group on self-reported social acceptance but there were no other significant differences.

While it is not possible to draw firm conclusions about the association between particular aspects of disfigurement and psychological functioning from this small group of studies, five of the nine studies that evaluated within disfigurement group differences found significant distinctions on different variables. As such, it is probably too early to exclude the possibility that disfigurement-related factors may have an impact on psychological functioning.

Gender and Age

Three papers compared boys and girls in their disfigured sample. All three found no main effects for gender but reported interaction effects. Leonard et al (1991) and Shute et al (2007) reported a significant interaction effect between age and gender with older girls fairing worse than younger girls in both studies on self-esteem and in the Shute study only on satisfaction with appearance. Leonard et al (1991) also found that older boys reported significantly better outcomes than younger boys. Shute et al (2007) did not find an age effect for boys. Both studies sampled only young people with oro-facial and cranio-facial anomalies but the age ranges of each

study were different with the Leonard study including 8-18 year olds and the Shute study focusing on 11 to 14 year olds.

The third study that investigated the impact of gender on psychological functioning in young people with disfigurement, Feragen et al (2010), also found no main effect based on gender, but reported an interaction effect between gender and visibility of disfigurement as well as finding a main effect for visibility of disfigurement on some outcome measures. Girls with a non-visible disfigurement were significantly more satisfied with their appearance than girls with a visible disfigurement and girls in the non-disfigured comparison group. Boys with a visible disfigurement, on the contrary, reported significantly better close friendships, social acceptance and depressive symptoms.

No main effects were found for any demographic variable tested. Interaction effects were found for gender and age, with older girls faring worse than younger girls and older boys doing better than younger boys. However, each of these findings were reported by a small number of quite dissimilar papers making it difficult to draw any conclusions with confidence.

1.1.5.2.6 Limitations of the Review.

Defining the population

The first limitation of this review relates to the definitional complexities of the term "disfigurement". First, because disfigurement is a descriptive term that can be applied to a number of physical manifestations, the populations being studied were quite heterogeneous; in terms of the nature of the disfigurement itself, as well as in terms of physical and functional co-morbidities. The largely descriptive studies that were included in this review were not able to account for much of this heterogeneity

within the disfigured population. Second, despite the fact that many of the studies measured disfigurement severity, because of the different types of disfigurement involved and the idiosyncratic ways in which studies measured disfigurement severity, it was impossible to determine if each population was comparable in terms of the social processes that are hypothesized to contribute to psychological dysfunction in disfigured groups. Similarly, the available data did not allow the partialling out of the effect of being disfigured from other effects of having the medical condition.

Sampling

There were a number of issues with the quality of the studies that, in themselves, may not have been problematic but meant that the results of this review were potentially skewed. First, all studies drew their participants from specialist medical clinics with high levels of expertise and experience in managing the disfiguring medical condition. This may mean that people with the same medical condition but who are not seen in specialist medical services are not represented in the study samples. This could potentially contribute to the under-representation of people with milder versions of the same condition, people who do not receive the same specialist, and often well-resourced, healthcare support or, depending on the nature of healthcare provision in the country in which the sample was selected, people from lower socio-economic levels, or other marginalized social groups, who cannot afford, or are unable to access, specialist medical care. Second, 10 of the 12 studies used participants with CLP. As such, it is unclear if the reported outcomes are generalisable to individuals who have been disfigured via means other than CLP. Third, the majority of the studies were conducted in the USA. Only one study was conducted in the UK. As such, it is unclear how generalisable the findings of these studies are to research and clinical practice conducted in the UK.

Furthermore, not all studies reported ethnicity and occupational data for their samples, making generalizing the findings more difficult.

1.1.5.3 Discussion.

The current literature review set out to fulfill one key aim: to find out how children and adolescents with congenital disfigurement compared with the non-disfigured population psychologically. The key finding was that no consistent, unequivocal patterns of psychological functioning have yet been identified. The majority of the overall findings reported no significant differences between disfigured and non-disfigured young people. Some studies also found significantly better functioning in the disfigured group compared to the general population. The studies that reported significant psychological difficulties in their disfigured samples tended to find these in the domains of social difficulties and internalizing problems. As such, the overriding message was that psychological and social difficulties should not be assumed in young people with a disfigured appearance.

The additional findings were also mixed with some studies reporting within group differences based on medical diagnosis, disfigurement severity and gender and age together and others reporting no differences in those domains. In combination with the mixed findings of the comparisons between disfigured and non-disfigured groups, these results appear to suggest that this population of young people is psychologically heterogeneous and that there may be a wide range of variables that influence psychological functioning in this population. That is, a main effect relationship between congenital disfigurement and psychological functioning is unlikely. Rather, a multi-factorial causal pathway appears more plausible.

The findings of the studies described in this review were limited by the small number of studies conducted on the subject of congenital disfigurement in middle childhood

and adolescence and the fact that all but two of the studies sampled young people with cleft lip and/or palate or cranio-facial anomalies. There was also a consistent trend for self-reported outcomes to be better than non-disfigured norms and better than parent-reported outcomes. Furthermore, while there was little support for disfigurement severity as a key differentiating variable, the majority of studies testing for within group differences found significant variation of some sort within their disfigurement sample. This suggests that disfigurement samples may be quite heterogeneous. While some within group differences had prima facie validity, such as older girls feeling more negatively about their appearance and more socially marginalised than their younger counterparts (Shute et al, 2007), others, such as older boys reporting better psychological outcomes than younger boys (Leonard et al., 1991) or more severely disfigured young people reporting better psychological functioning than mildly disfigured young people (Andersson et al, 2012) are still slightly perplexing.

Overall, the combined results of the papers did not create a coherent picture of psychological functioning in children and young people with congenital disfigurement. The findings are equivocal and heterogenous, with significant methodological limitations characterizing much of the area, suggesting that there is scope for further research into congenital disfigurement in childhood and adolescence. Furthermore, the specific process by which disfigurement has a differential impact on those affected by congenitally disfiguring medical conditions is unclear due to a lack of relevant research despite the existing studies indicating the congenitally disfigured population to be quite psychologically heterogeneous.

1.1.6 Understanding Psychological Heterogeneity in Young People with Congenital Dermatological Disfigurement

The clearest finding of the current literature review is that there appears to be a wide range of psychological and social functioning reported in children and adolescents with congenital disfigurement ranging from better than normal to worse than normal. The empirical literature on disfigured adults reports a similar mixed picture of psychological functioning from significantly better than the general population to significantly worse than the general population (e.g., Rumsey et al., 2004; van den Elzen et al., 2004). This heterogeneity suggests that a disfigured appearance does not have a main effect on psychological functioning. Rather, it suggests that there may be a number of variables, within each affected individual, that influence the psychological and social impact of the disfigurement. This is in keeping with current general models of psychological development, such as the developmental psychopathology model (e.g., Cicchetti & Toth, 1997)

Researchers in the disfigurement field have responded to the psychological heterogeneity evident in this population by postulating the need for multivariate explanatory models to explain this heterogeneity within the disfigurement population. Rumsey et al (2004) commented on the importance of developing a multi-factorial model to understand appearance-related concerns. Thompson (2012) argued that there was a paucity of theoretical development in understanding the role of appearance in psychological adaptation emphasizing the importance of linking theory and method to practice. Thompson emphasized the importance of developing models, theories or frameworks to explain psychological functioning that ultimately lead directly to clinical intervention planning, that is, with direct translational value. Thompson further argued the importance of balancing disfigurement-specific variables with what is known about psychological commonalities.

Explanatory Model of Psychological Functioning in Adults with Disfigurement

In the largest study of its kind to date, the Appearance Research Consortium (ARC)(Clarke et al., 2013) conducted a nationwide, multi-site study with adults with self-defined disfigurement. The study aimed to address two key issues, which the research team viewed as significant gaps in the existing literature. The first concerned sample difficulties. The research group believed that existing studies on this population had been weakened by small and disparate samples and by the fact that most studies drew their samples from specialist hospital settings, thereby excluding the large group of affected individuals who did not receive specialist healthcare support for their disfiguring condition. The second was the lack of clear findings on factors that influenced psychological functioning in this population. Earlier papers such as the Fajervik-Morton (2008) systematic review concluded that, while a small number of studies had investigated a range of possible contributing factors, the evidence was still very limited. The lack of evidence regarding contributing factors was seen as key to the research group due to the dissonance between the nature of the disfigurement and the psychological functioning of the individual, which characterizes this population. The researchers also highlighted the importance of ensuring the clinical relevance of the findings in order that effective interventions could be driven directly from the findings of the study, which was, in part, the reasoning behind the team's focus on mediating cognitive processes that might account for the discontinuity between the physical features of the disfigurement and psychological functioning.

While there were several components within the whole research programme, the largest part of the project involved testing a number of variables thought to significantly influence the outcomes of appearance-related concern and psychological well-being. The variables had all received research attention in the appearance literature and had some empirical support. The team acknowledged

that they had used a pragmatic approach, rather than a theoretically driven approach, in identifying factors to test in their model. That is, factors that had already found empirical support.

The explanatory model that provided a framework for the ARC study of contributing factors was first described in a book chapter authored by Thompson (2012) and consisted of three parts. The first part contained the predisposing variables that the team believed to be relevant in explaining psychological functioning in this population but which were not accessible to psychological intervention. These included disfigurement-related factors, such as visibility of the disfigurement, demographic factors such as gender, and socio-cultural influences such as media, family and peer influences. The second part contained the cognitive processing variables including dispositional style, perceived social support, fear of negative evaluation and salience and valence of appearance. The third and final part of the model included the psychological "outcome" variables of appearance-related concerns and indices of psychological well-being. The research group acknowledged that the relationships between these groupings and individual variables were not considered. They also acknowledged the complexities of placing individual variables into categorical groups when certain variables could easily belong in one or more groupings.

Results showed strong support for the explanatory model, which was based on a five-block regression model. The largest amount of variance explained was in the outcome of appearance anxiety (R^2= 0.662, F(16, 1038)=127.191, MSE=85.91, p<.001. Approximately 50% of the variance was explained in the outcomes of general anxiety, depression and negative affect and 20-30% of the variance was explained in the remaining variables of aggression and positive affect. The high amount of variance explained in appearance concerns is not surprising given the

similarity between the outcome variable and some of the predictor variables. However, the moderate amount of variance explained in the general mental health indices is compelling.

There were a number of limitations of the ARC study that affect its usefulness for understanding children with congenital disfigurement. This model garnered strong empirical support for the association between its component variables. However, the causal relationships between each stage of the model are still unclear. First, the model was of an additive, empirically-based nature rather than causally or theoretically coherent. For example, while age was found to be indirectly correlated with negative social functioning, the link between these two factors was not easily explained. Second, all measures of disfigurement and psychological functioning were self-rated. As a largely cross-sectional study, it was impossible to determine whether disfigurement influenced psychological functioning ratings or if, in fact, psychological functioning influenced disfigurement ratings. For example, the design of the study was such that it was impossible to determine the presence of major psychological disorders such as Body Dysmorphia, that may have led a participant to describe themselves as disfigured and even in the presence of a disfiguring medical condition it was not possible to corroborate the participant's ratings of their disfigurement. Third, shared method variance may have accounted for some of the correlations between independent and dependent variables as all data were reported by the participant alone. Fourth, it is unclear whether this model, tested on adults and based on the adult literature, has any relevance to children and adolescents. It is generally considered to be unsafe to extrapolate models of adult psychological functioning to children without question, particularly in the case of cognitive processes. The development, role and stability of cognition in psychological functioning and psychological therapies for young people is still questionable. As such, the main focus of the ARC model, cognitive processing, may

not be valid, relevant nor clinically useful in the field of childhood adaptation to disfigurement.

1.1.7 The current study

The current study was driven by the findings of the review of the literature as well as the recent work that has been attempting to account for the psychological heterogeneity in the adult disfigurement population (Clarke et al, 2013). The current study aimed to further the existing literature on psychological functioning in young people with congenital disfigurement by addressing weaknesses in the current knowledge-base identified in the described literature review as well as considering a range of variables that might contribute to the psychological heterogeneity in this population.

The current study aimed to address the limitations in the existing literature by:

1) Limiting the indices of psychological functioning to just two domains: social and emotional functioning as there is some evidence for difficulties in these domains in the disfigured population.

2) Investigating school-aged children and adolescents rather than adults with congenital disfigurement.

3) Investigating congenital dermatological disfigurement rather than cleft lip and/or palate as this particular type of disfigurement is significantly under-researched.

4) Utilising a large UK sample of young people due to the lack of sufficiently powered, UK-based studies on which to drive clinical developments relevant to the local population.

5) Using positive and negative outcome measures as the existing literature indicates that young people with congenital disfigurement may report better than normal functioning in the psychological domain as well as worse than normal functioning.

The main part of the current study aimed to understand the psychological heterogeneity within congenitally disfigured young people by investigating the impact of specific psychological, demographic and medical factors on socio-emotional functioning within the dermatologically disfigured sample. In particular, this study aimed to:

1) Investigate the impact of gender and developmental stage on socio-emotional functioning. Studies reviewed found weakly positive support for the role of gender (Feragen et al, 2010; Leonard et al, 1991; Shute et al, 2007) and developmental stage (Leonard et al., 1991; Shute et al., 2007) on psychosocial functioning in the congenitally disfigured population.

2) Investigate the impact of co-morbid medical symptoms on socio-emotional functioning. One study that investigated psychosocial differences between diagnostic groups (Sheerin et al, 1995) reported significant differences between the group with port-wine stains and the group with prominent ears while another (Kapp-Simon et al., 1992) reported no difference between a cranio-facial

group and a cleft lip and palate group. Due to the lack of research on the impact of the medical diagnosis on psychosocial functioning, and the mixed findings of the reviewed studies, the current study will investigate whether medical diagnosis differentiates socio-emotional functioning in the congenital dermatological disfigurement group. In addition, a recent paper that did not fit the inclusion and exclusion criteria for the current literature review by Feragen, Stock & Rumsey (2014) reported the significant impact of medical and developmental co-morbidities on the psychological functioning of a cleft lip and palate sample. As such, the medical comorbidities commonly associated with young people with congenital dermatological disfigurement will also be investigated to determine if the presence of symptoms other than disfigurement differentiates socio-emotional functioning with the sample.

3) Investigate the role of two psychological factors known to influence socio-emotional functioning in the general population: attachment and shame. These two variables have received limited attention in the field of disfigurement research with mixed results. The rationale for considering attachment and shame in the aim to understand the heterogeneity in disfigured young people will be explained in Section 1.2.2.1 and Section 1.2.2.2.

1.2 Section 2: Socio-Emotional Heterogeneity and Congenital Disfigurement

In order to inform empirical but especially clinical developments to support young people towards an adaptive developmental trajectory, it is important for researchers to try to identify the variables most likely to affect, or that have the greatest impact on, the people who have experienced a particular risk factor. Understanding what

facilitates positive psychological adaptation as well as what contributes to psychological maladaptation will help to identify the key factors on which to focus psychological interventions or the factors that might identify a young person as being at risk of developing psychological difficulties.

The following section:

1) Describes the developmental framework for this study.
2) Discusses disfigurement within the developmental framework.
3) Describes the two hypothesised contributing variables, attachment and shame, and provides the rationale for their proposed role in influencing socio-emotional functioning in young people with congenital disfigurement.

1.2.1 Psychological Development

Many questions remain about psychological functioning in children and adolescents with congenital disfigurement. However, one phenomenon is emerging in the current literature: congenital disfigurement, like many other risk factors, appears to have a differential impact in individuals. While some people experience major psychological and social difficulties related to their disfigurement, many others experience no difficulties at all and some report highly successful and satisfying lives.

1.2.1.1 Understanding psychological development.

The findings of the review suggested that the socio-emotional impact of congenital disfigurement is complex and unlikely to take the form of a simple, main effect relationship. While studies have begun to test the effect of moderating factors on the relationship between disfigurement and psychosocial function, few have linked the extensive literature on general child psychological development with the disfigurement literature to form psychologically coherent multi-factorial causal

pathways that might explain the varied adjustment to disfigurement reflected in the descriptive studies so far.

The current study postulated that congenital disfigurement acts as a risk factor for social and emotional difficulties via the following pathway. Because physical appearance plays an important role in social roles and relationships, congenital disfigurement increases the risk of experiencing social rejection or devaluation. Social rejection might take the form of implicit devaluation or marginalisation such as in the case of stigma, explicit social rejection such as teasing and bullying or ambiguous but intrusive social experiences such as staring. Social rejection and devaluation act as psychological stressors and increase the likelihood of the individual experiencing significant socio-emotional problems. The impact of this social stress on socio-emotional functioning will be mediated by other factors that influence stress-coping.

There are many well-established psychological models that help us to understand the differential impact of stressors on individuals. Because this study focused on the developmental stages of middle childhood and adolescents, the Developmental Psychopathology Model (Rutter & Sroufe, 2000) was used as a framework by which to understand heterogeneous socio-emotional functioning within the congenitally disfigured population.

1.2.1.2 The role of physical appearance in the social context.

"Beauty is good" is an evolutionary theory tested in a seminal paper by Dion, Berscheid & Walster (1972). In the study, 30 male and 30 female participants were shown photographs of three stimulus people who had previously been ranked by 100 university students as attractive, moderately attractive or relatively unattractive. The participant was asked to make a series of ratings and projections about the

stimulus people that were then grouped into seven dimensions: social desirability, occupational status, marital competence, parental competence, social and professional happiness, total happiness and likelihood of marriage. The results indicated that the attractive stimulus person was rated most positively and the unattractive stimulus person least positively on six of the seven domains, the exception being the domain of parental competence. These results were reported as indicating significant differences by the authors. The gender of the participant and the stimulus person were controlled for and no effect was found for the interaction of genders. The attractiveness of the people represented in the test stimuli was also rated by the participants and these attractiveness ratings were consistent with those of the pre-experiment ratings.

Further evidence to support the findings of Dion et al. (1972) has been plentiful such as reports that attractive people, on average, earn 12% higher incomes than unattractive people (Hamermesh & Biddle, 1993) and that people of all ages, including infants, demonstrate preferences and biases in favour of attractive people (e.g., Langlois et al., 2000b). It has been hypothesised that a tendency to attach positive attributes to physically attractive individuals has its basis in the basic evolutionary drives to survive and to pro-create (e.g.,Tovée, Maisey, Emery, & Cornelissen, 1999). A series of meta-analyses of studies testing the beauty is good theory was published by Langlois, Kalakanis, Rubenstein, Larson, Hallam, & Smoot in 2000. Their findings unequivocally supported the notion that attractive people were more likely to be imbued with positive psychological and social attributes. The study also found that this finding was consistent across children and adults.

Griffin & Langlois (2006) furthered the empirical understanding of the “Beauty is Good” theory when they demonstrated that it was not just that beauty is good but that also, “ugly is bad”. In experiments that studied each adult and child respondents

separately, unattractive people were the recipients of a negative bias compared to people who were of average or high attractiveness, in a similar way that attractive people were more likely to have a positive bias.

In conclusion, there is a substantial body of empirical research that suggests that individuals with a disfigured appearance are more likely to be treated in a negative, devaluing and rejecting way than individuals of average or high attractiveness.

Implicit Social Rejection

There has been some limited research to support the hypothesis that disfigured people experience social rejection. In the best known experimental study of social reactions to a disfigured person, Houston & Bull (1994), compared the reactions of passengers on a train to a confederate without a visible difference to the same confederate cosmetically made up to show a facial port-wine stain. When busy-ness of the train carriage and number of seats available were controlled for, significantly fewer people sat near to the confederate with the port-wine stain than the same confederate without a disfigurement.

A number of recent experimental studies have investigated the impact of a disfigurement on the information processing of the observer. The results of these studies have found, consistently, that having a disfigured appearance has a negative impact on the information processing of the observer. For example, Madera & Hebl (2012) conducted two studies. Over one thousand participants were shown a recorded job interview with either a facially disfigured or non-disfigured applicant. The results showed that participants watching the disfigured applicant spent significantly more time looking at the body area on which the disfigurement was located, they remembered less of the interview content and they rated the disfigured applicant significantly lower than the non-disfigured applicant. In the

second study, 38 managers conducted a face-to-face interview with either the disfigured or non-disfigured applicant. Again, the managers who interviewed the disfigured applicant recalled less of the interview content and rated the applicant lower than the non-disfigured counterpart. The managers who interviewed the disfigured applicant were also found to deplete more self-regulatory resources as measured by a simultaneously run Stroop Task (Macleod, 1991).

In a similar study, Stevenage & Furness (2008) found that conversational recall dropped significantly when the stimulus job applicant was disfigured. However, this study also found that there was no difference in personality ratings between the disfigured and non-disfigured applicants.

A large scale project reported by Richardson (1983) and continued by Harper (e.g., 1995; Harper & Peterson, 2001) evaluated social preferences in children between the ages of six and 16 across several cultural groups including a mixed American population, New Zealand Maori, Nepalese and Antiguan. Children were presented with pictorial images of a non-disabled child, a child with a full leg brace and crutches, a child in a wheelchair, a child with a cleft lip and an obese child. The images were controlled for gender and ethnicity.

Consistently, the most socially preferred stimulus child was the non-disabled child while the least preferred stimulus children were the facially disfigured child and the obese child. This effect was more powerful in the US sample although significant for all ethnic samples (Harper & Peterson, 2001). There was some variation across samples with some non-Western samples more likely to rate the obese child more positively and more likely to rate the physically disabled child more negatively.

Explicit Social Rejection

In addition to studies on these internal processes linked with social rejection, evidence of explicit social rejection has been found in adults with a visible difference. Strauss, Ramsey, Edwards, Topolski, Kapp-Simon, Thomas, Fenson & Patrick (2007) reported, in a study on adults with port-wine stains (PWS), that 75% of participants reported that people stared at them because of their PWS and 73% reported that they had felt hurt by what other people had said in relation to their PWS. Similarly, 35% of adolescents aged 11 to 18 with facial disfigurement reported that they had experienced others staring, 29% reported comments made to them about their disfigurement and 20% had been teased because of the disfigurement in the past month. Teasing and bullying experiences have been shown to be negatively associated with mental health (e.g., Hunt, Burden, Hepper, Stevenson, & Johnston, 2007; Rimmer, Foster, Bay, Floros, Rutter, Bosch, Wadsworth, & Caruso, 2007). In a recent study by Feragen & Borge (2010), the authors concluded that their sample of children and young people with visible and non-visible cleft reported a relatively low level of teasing by peers (25-41%) compared to general population estimates and previous studies on young people with CLP such as that of Turner et al. (1997) who reported teasing in 60% of their sample. However, the authors found a significant association between peer harassment and dissatisfaction with appearance, highlighting the influence of socially rejecting experiences on this group of young people. However, as a non-disfigured control group was not employed in these studies, it is difficult to determine whether the level of perceived social rejection was greater in the disfigured population than in a non-disfigured population. Research has found self-reported levels of teasing as high as 75% in adolescents who are not disfigured (Lovegrove & Rumsey, 2005). Furthermore, because these studies were based on self-report, it is unclear how much of these results are attributable to the disfigured individuals' internal biases.

Conclusion

In summary, research has found compelling support for the hypothesis that social judgements are driven by implicit processes in which attractive individuals are judged more positively and unattractive individuals are judged less positively (e.g., Langlois et al., 2000). There is further evidence to suggest that these processes can impact on decision-making and behaviour in a way that is prejudicial to the individual with a disfigured appearance (e.g., Houston & Bull, 1994, Madera & Hebl, 2012) As such, there is substantial support for the hypothesis that having a disfigured appearance can result in negative social experiences constituting a chronic social stressor. The stress literature suggests that experience of increased levels of stress has been shown to be associated with poor psychological outcomes (e.g., Grant et al, 2003). However, as argued by the developmental psychopathology model, stressors are unlikely to have a direct effect on psychological functioning (Grant et al, 2003). Rather, there will be a complex pathway of contributing variables that determine whether the stressor affects the individual's psychological functioning, thereby accounting for the high degree of variation in psychological function in the disfigured population.

Sections 1.2.2.1 and 1.2.2.2 of this chapter describe the two developmental variables, attachment and shame, that this study hypothesized might influence the relationship between disfigurement and the individual's socio-emotional functioning.

1.2.2 Factors contributing to socio-emotional heterogeneity.

The current study set out to test a multi-factorial developmental framework to account for socio-emotional heterogeneity in children and adolescents with congenital dermatological disfigurement. The model postulated that the way in which congenitally disfigured young people respond to social stress would be influenced by two factors: attachment and shame.

Both variables were selected from a wide range of potential contributing variables for the following reasons, which will be explained in more detail in Sections 1.2.2.1 and 1.2.2.2:

1) Both factors have been empirically shown to be associated with psychological outcomes in non-disfigured populations (e.g., Gilbert, 2000; Sroufe, 2005).

2) Both factors have been associated, in theory and empirical research, with social functioning, cognitions, emotional responses and social behaviours (e.g., Bowlby, 2004; Gilbert, 1993).

3) Both factors form part of the transactional process that contributes to how the individual interprets and responds to social experiences which subsequently contributes to social interactions becoming more or less successful (e.g., Bowlby, 2004).

4) Both factors have been shown to be amenable to therapeutic change. As such, they could potentially drive developments in clinical practice for helping children and young people with congenital disfigurement to achieve better socio-emotional functioning (e.g., Bakermans-Kranenburg, Van Ijzendoorn, & Juffer, 2003; Gilbert, 2009).

5) Both factors have been linked to one another and might work, in a combined way, to influence socio-emotional outcomes (e.g., Schore, 1998).

1.2.2.1 Attachment

Attachment is a well-known construct in child clinical psychology and one of the original psychological factors to be specifically located in a developmental pathways framework (e.g., Bowlby, 1988). There is a large body of literature that links attachment style to a range of indices of psychological functioning across the lifespan (e.g., Grossmann, Grossmann, & Waters, 2005).

In the current study, attachment was hypothesised to be associated with the socio-emotional functioning of children and adolescents with congenital disfigurement for two specific reasons. First, disfigurement was hypothesized to create psychological stress by negatively impacting on social interactions.

Attachment, which is often conceptualized as a template for all social interactions and relationships, was hypothesized to ameliorate negative social experiences relating to the young person's disfigured appearance Attachment security would increase the likelihood of positive social experiences and expectations and decrease the likelihood of negative social experiences as well as influencing the way in which the individual understands and responds to negative social interactions thereby mitigating the impact of social rejection or devaluation.

Second, based on early studies on disfigurement in infancy, it was hypothesized that disfigurement could act as a barrier to the formation of secure attachment between parents and their babies due to, for example, the parent being less responsive to their infant because of their ambivalent feelings about the baby's disfigured appearance (e.g., Wasserman & Allen, 1985).

Therefore, congenitally disfigured children were hypothesised to be more likely to be insecurely attached due to the impact of disfigurement on attachment formation and that their attachment insecurity would increase the risk of negative social experiences due to the impact of their attachment status on their own social behaviours and cognitions. The details of these two proposed causal pathways are detailed below.

1.2.2.1.1 Background.

According to the progenitor of attachment theory, John Bowlby, the attachment system is a set of threat-oriented proximity seeking behavioural systems directed towards the primary caregiver that are designed to increase an infant's chances of survival by protection from threat and facilitation of safe exploratory behaviour. Bowlby theorized that, as the infant develops to adulthood, the same behavioural constellation becomes more complex and sophisticated in order to achieve "inclusive fitness" within one's social grouping rather than with just the primary caregiver. The individual applies the implicit threat-based behavioural routines, already learnt in infancy, to drive this more complex network of sustained, threat-reducing, life-enhancing relationships (Bowlby, 1988).

The concept of attachment emerged from Bowlby's work with anti-social adolescents (e.g., Bowlby, 1950; 1951) during which he noticed the association between maternal deprivation and the absence of affection, shame and a sense of responsibility in this group of adolescents. Bowlby formed conceptual links between his observations and those of his contemporaries such as Konrad Lorenz and his ethological perspective on mother-infant interactions (e.g., Lorenz, 1971). Bowlby postulated that humans had an innate and universal need to form close affectional bonds and that an infant formed a close bond or dependence with a "secure base", in which attachment was seen to be the behavioural and motivational system that facilitated physical and psychological proximity to the caregiver. Bowlby formed the idea of the attachment figure as the source of security and support from whom the infant felt safe and motivated to explore the outside world. Attachment not only described the child's need to be close to its attachment figure but also explained how this feeling of security promoted the child's competent exploration away from the attachment figure (Bowlby, 1977b).

Based on these fundamental premises, Mary Ainsworth (Ainsworth, Bell, & Stayton, 1971) developed the attachment behavioural system construct by operationalising the notion of maternal sensitivity to infant signals and its role in the development of idiosyncratic patterns of interaction between each mother and infant. The range of behavioural patterns that develop in response to early experiences with the primary caregiver have since been well described and characterised using various observational, interview, and questionnaire methods. Attachment behaviours have broadly been found to consistently cluster into types; those based on the expectation of a safe environment, and those developed to respond to an environment perceived as threatening. Behaviour patterns developed in the safe environment are described as a secure attachment style, whereas those learnt in the context of a threatening environment are referred to as insecure attachment style (Ainsworth et al., 1971).

1.2.2.1.2 Attachment categorization.

Mary Ainsworth and her research team were the first to document that, on triggering an infant's attachment behaviour, there were striking individual differences in infant responses (Ainsworth et al., 1971). The process of eliciting attachment behaviours under experimental conditions developed into what is now considered one of the gold-standards of attachment measurement in infancy: The Strange Situation (Ainsworth, Blehar, Waters, & Wall, 1978). These differential patterns of infant behaviour were then linked to specific patterns of interaction between the mother and the infant in the year preceding the observation and led the authors to conclude that, based on lived experience of maternal responses, the infant formed internal models of effective proximity-seeking behaviour that were being elicited in stressful situations, such as in the Strange Situation. Initially, Ainsworth and colleagues identified three patterns of attachment behaviour; secure, insecure-avoidant and

insecure-ambivalent/resistant. A fourth category, referred to as disorganised or disoriented was added later (Main & Solomon, 1986).

Secure Attachment (B)

Secure attachment style, also referred to as Group B, is characterized by an infant who, in Ainsworth's Strange Situation, demonstrated a willingness to explore the new (experimental) surroundings in the presence of their mother, showed signs of missing their mother when she left the room, was not placated by the presence of a stranger during the mother's absence, but was reassured and calmed on reunion with the mother. Studies consistently demonstrated greater maternal sensitivity in the mothers of secure infants than in the mothers of insecure infants (Bretherton, 1985). Current estimates indicate that approximately 60-66% of the UK population are in the secure attachment category (e.g., Ainsworth et al., 1978; Shmueli-Goetz, Target, Fonagy, & Datta, 2008; Van Ijzendoorn & Kroonenberg, 1988; Van Ijzendoorn, Schuengel, & Bakermans-Kranenburg, 1999).

Insecure-Avoidant (A)

Insecure avoidant attachment style, also referred to as Group A, is characterized by infants who showed little distress throughout the Strange Situation procedure during separations and reunions and, on the mother's return, avoided or ignored their mother, appearing to be more interested in other things such as the toys or other features of the room. Despite the apparent lack of distress, later studies consistently found that these children were actually experiencing physiological signs of distress akin to the secure infants, if not even higher levels, leading the authors to conclude that insecure-avoidant infants suppressed the behavioural expression of their distress on separation (Fraley, Davis, & Shaver, 1998). Studies have also noted that the physiological arousal in this group of children, elicited by separation, also took longer to subside on reunification with their parents, despite the lack of behavioural

signs of distress (Spangler & Grossmann, 1993). The Ainsworth team linked this group of children to maternal rejection of attachment behaviours. As a result, the authors surmised that these infants learnt to refrain from expressing distress and any other forms of attachment behaviour by shifting their attention from their mother or the absence of their mother toward the inanimate environment. According to the large-scale studies on attachment classification, 15-28% of the population can be categorized into Group A (e.g., Ainsworth, 1978; van Ijzendoorn & Kroonenberg, 1988; van Ijzendoorn et al., 1999; Shmueli-Goetz, Target, Fonagy & Datta, 2008).

Insecure-Ambivalent/Resistant (C)

Insecure-Resistant/Ambivalent attachment, also known as Group C, is characterized by heightened emotionality throughout the Strange Situation process including marked fear, in some infants, on entering the experimental room despite their mother's presence, little exploration of the room and an inability to be settled by their mother. On reunion with their mother, infants expressed a mixture of subtle to open resistance towards their mother with continuing, simultaneous expressions of distress and proximity-seeking. The authors hypothesized that the experience of unpredictable responses to proximity-seeking behaviours meant that infants needed to amplify their proximity-seeking overtures, even in safe situations, to increase the likelihood of eliciting a response from their caregiver (Main, 1990). Studies estimate that 3-15% of the population can be categorized into Group C (e.g., Ainsworth, 1978; van Ijzendoorn & Kroonenberg, 1988; van Ijzendoorn et al., 1999; Schmueli-Goetz, Target, Fonagy & Datta, 2008).

Disorganised/Disoriented (D)

This categorisation was created later on when researchers found that there was a group of infants who could not be easily categorized into the initial three groups. These infants displayed no apparent consistent pattern of attachment behaviour,

unlike the previous groups where the children had apparently adapted coherent styles of behaviour to fit with their mother's style of interaction. The authors believed that this collapse of coherent behavioural and attentional strategies occurred when the infant was frightened by the attachment figure, where the innate drive to seek comfort from the parental figure when frightened was met with frightened, frightening or dissociated behaviour from the parent. Studies have reported incidence levels of 4%-15% of the population (e.g., van Ijzendoorn et al., 1999; Shmueli-Goetz, Target, Fonagy & Datta, 2008).

As a result of the early research conducted by Mary Ainsworth and her colleagues, attachment style in infancy can now be fairly reliably categorized into the four attachment groups; secure, insecure-avoidant, insecure-ambivalent/resistant and disorganized. There have been some alternative means of categorizing attachment style proposed over time such as Crittenden's spectrum-based approach (Crittenden, 2000) and subtle variants on Ainsworth's categorizations such as those used in the Adult Attachment Interview (George, Kaplan, & Main, 1984; 1985; 1996) and Child Attachment Interview (Shmueli-Goetz et al., 2008). However, Ainsworth's original categories have remained predominant and have repeatedly been shown to be reliable and valid (Ainsworth, Blehar, Waters, & Wall, 2014).

1.2.2.1.3 Attachment and development

Researchers have argued for five specific ways in which attachment style might uniquely affect development and psychopathology. The first causal mechanism, espoused by Bowlby (2004), suggested that attachment style, based on early experiences of parent-infant interaction, formed the basis of fundamental models of the self and interpersonal relationships known as internal working models (IWMs). Cognitive affective structures or IWMs of self and self in relation to others would thereby contribute to the securely attached individual for example, seeing

themselves as competent and valued and the relational world as warm, reciprocal, responsive and trustworthy. In contrast, insecure attachment may lay down IWMs that are characterized by helplessness, mistrust, isolation or fear (Main, 1995). These different IWMs fit well with cognitive attributional bias models in children and adults with internalizing and externalizing difficulties (e.g., Farina, Leifer, & Chasnoff, 2004; Manassis, 2001).

The second mechanism by which attachment may affect development is via the process of affect regulation. Schore (1998) argued that active parental participation in regulating the infant's negative emotional state is critical in enabling the child to shift from the negative affective state of deflation and distress to a re-established state of positive affect. If the caregiver is sensitive, responsive and approachable, they will notice signs of the infant's internal distress and will naturally reinitiate an appropriate, affect-regulating interaction Without a responsive and sensitive attachment figure, negative emotional states and their neurobiological concomitants become prolonged and result in persistent states of heightened emotional stress. The infant does not learn how to regulate this affective state without the experience of external emotional regulation provided by the primary attachment figure. Emotional dysregulation has been associated with psychosocial maladjustment across the lifespan including social difficulties in preschool aged children (e.g., Rubin, Cheah, & Fox, 2001) and with major mental health problems in adulthood such as borderline personality disorder (e.g., Gratz, Rosenthal, Tull, & Lejeuez, 2006) and generalized anxiety disorder (e.g., Mennin, Heimberg, Turk, & Fresco, 2005).

The third mechanism is a behavioural link whereby undesirable behaviours such as whining or aggression are reinforced as effective proximity seeking behaviours in infants and young children, especially when adaptive proximity-seeking behaviours

have been ineffective or unavailable to the infant. These undesirable behaviours tend also to have the paradoxical effect of pushing potential caregivers away, thereby increasing relational anxiety in the infant causing them to utilize their undesirable strategies even more. In contrast, as indicated by several studies, securely attached children exhibit more effective, sophisticated and socially desirable proximity-seeking behaviours that ultimately reinforce the secure child's view of the world as a supportive, responsive environment and themselves as competent agents within that world (Biesecker, 2001; Cohn, 1990; Elicker, Englund, & Sroufe, 1992)

The fourth mechanism relates to motivation to engage in social interaction. Early attachment relationships that were rewarding in their warmth and responsiveness are likely to create a state of readiness and willingness to socialize. Children with positive experiences of social interaction might be more likely to seek out other social relationships and respond constructively to positive social overtures by the other person. Unresponsive or unpleasant early relationships may establish an expectation for all social relationships to be difficult and unhelpful which would likely deter children from seeking out new relationships or maintaining existing relationships (Greenberg, 2002).

The fifth mechanism is that secure attachment contributes to an infant's development of reflective capacity; the ability to adopt a meta-stance in relation to their own internal world and the external world. Fonagy & Target (1997) argued that the relationship between attachment and later psychosocial function was mediated by the development of reflective capacity and subsequently mentalization; the ability to "mind-read". The ability to mentalize, or to form accurate hypotheses about what other people are thinking and feeling facilitates the construction of positive relationships and inhibits malevolent or anti-social acts (Fonagy & Target, 1997). In

contrast, in the absence of consistent, mirroring and attuned interactions and without the experience of repeated interrelational rupture and repair with early attachment figures the infant is unable to develop the requisite understanding of how their own internal world relates to other minds and the external world.

1.2.2.1.4 Concurrent attachment and socio-emotional functioning.

The majority of attachment research in childhood has focused on attachment in infancy. More recently, there has been more research interest in concurrent attachment and psychological functioning in adults i.e., attachment style at the time of measuring other psychological variables. Much less research has been conducted on concurrent attachment in children beyond infancy and up to adolescence.

While attachment theory suggests that attachment style remains stable over time at the levels of cognitive, behavioural and emotional processing (e.g., Bowlby, 1977a) and empirical research has supported the association between infant attachment and psychological functioning in later life, there is emerging evidence to indicate a significant degree of instability in attachment style over time (e.g., Weinfield, Sroufe, & Egeland, 2000). As such, infant attachment style may have a limited predictive value in relation to later psychological functioning. Concurrent attachment, however, has been demonstrated to have a higher association with psychological functioning in adulthood and adolescence (e.g., Adam, Sheldon-Keller, & West, 1996). However, concurrent attachment is complex, particularly in an empirical context, due to difficulties in determining causal direction in the relationship between attachment status and other psychological factors.

Attachment stability over time.

There have been a number of large-scale longitudinal studies of attachment that have provided some evidence regarding the stability of attachment over time. In a longitudinal study, Hamilton (2000) used the Strange Situation to measure attachment in infancy and the Adult Attachment Interview (AAI)(George et al., 1996) to measure attachment in adolescence in the same participants. The study found that 77% of secure-insecure classifications remained stable between infancy and adolescence.

Waters, Merrick, Treboux, Crowell & Albersheim (2000a) in an associated study found 72% consistency in the secure-insecure classification between infancy and young adulthood. Both studies found that the key reasons for discontinuity were significant life events such as loss of a parent through death or divorce, a parent or the child contracting a life-threatening illness, parental mental health problems or physical/sexual abuse of the child. Waters et al. (2000a) reported a 78% concordance between Strange Situation and AAI ratings in participants who had not experienced any major life events, although individuals who had experienced major life events had a much poorer concordance of 44%.

These findings based on longitudinal, epidemiological studies are supported by studies that have specifically measured attachment before and immediately after life changes and that have also observed significant changes in attachment style such as after the birth of a sibling (e.g., Teti, Gelfand, Messinger, & Isabella, 1995). A study by Weinfield et al. (2000) followed up the infants of high-risk mothers. The mothers' high-risk status was defined by the young age of the mother, single-parent status, low educational level, income at or below poverty level, unplanned pregnancy and self-reported high stress and low social support. The authors found a much lower rate of attachment stability between infancy and late adolescence of

only 38.6%. While initially appearing to contradict the findings of their earlier studies, the authors concluded that their findings were, in fact, consistent with previous studies in that the high-risk families simply seemed to experience far higher rates of negative life events, which correlated with their lower rates of attachment stability. The researchers noted that those individuals who demonstrated continuity of attachment style over time also appeared to have remained in stable family settings. They hypothesised that adolescent and adult attachment may not be due to an internalized working model of social relationships but a representation of parenting received. These authors questioned whether there is any internal stability in attachment at all. Rather, that it may be an epiphenomenon representing a relatively stable pattern of attachment behaviour from the parental figures which was facilitated by a stable environmental context.

Another source of support for attachment discontinuity over time is the fact that purposeful intervention has been found to be successful in changing insecure attachment to secure attachment such as in the case of psychological intervention (e.g., Bakermans-Kranenburg et al., 2003) and when abandoned or maltreated children have been adopted or fostered (e.g., Smyke et al., 2010).

In conclusion, the existing research suggests that attachment style is largely stable over the lifespan but is sensitive to significant life changes. This phenomenon is sometimes referred to as "lawful discontinuity". This balance of stability and instability means that the link between infant attachment studies and concurrent attachment studies is significant but not simple, especially in the cases of the most vulnerable young people who are more likely to have experienced major life stressors. Factors contributing to psychological difficulties, such as significant disruption to family relationships, may affect psychological function and attachment

style separately or affect attachment via changes in other areas of psychological function.

Concurrent attachment style and psychological functioning.

In possibly the most influential longitudinal study of attachment and development, The Minnesota Parent-Child Project Sroufe (2005), one of very few studies with sufficiently long follow-up, showed that both early and concurrent attachment security were associated with psychopathology reflecting some degree of stability over time. The study also found that the closer the measurement of attachment in time to the psychological outcomes measurement, the higher the association, suggesting a gradual deterioration over time in the relationship between attachment style and psychological functioning. This finding would fit with the Waters group theory of negative life event impact, in that the more time passes, the more chances there are for the occurrence of negative life events that might affect attachment style (Sroufe, Egeland, Carlson, & Collins, 2005).

Relatively few studies have examined the association between concurrent attachment and psychological functioning in middle childhood. Children with disorganized attachment styles have been described as standing out as demonstrating more aggressive behaviour than their peers while ambivalent attachment style was also associated with externalizing behaviour in boys and controlling, dependent behaviour in girls (e.g., Moss, Smolla, Cyr, Dubois-Comtois, Mazzarello, & Berthiaume, 2006; Turner, 1991). However, the evidence for an association between other attachment categories and psychological functioning is yet to be established.

There have been many more studies focusing on the association between concurrent attachment style and psychological functioning in adolescence. Insecure-

pre-occupied adolescents have been repeatedly found to be significantly more likely to present with depression and suicidality (e.g., Adam et al., 1996; Allen, Moore, Kuperminc, & Bell, 1998). Dismissing adolescents, on the other hand, seemed more likely to exhibit "externalizing behaviours", such as substance misuse (Allen et al., 1998). Becker-Stoll & Fremmer-Bombik (1997) found that dismissing adolescents showed the least autonomy and relatedness in interactions with parents. However, because of high levels of co-morbid and diffuse emotional and behaviour problems often reported in insecure adolescents, researchers have hypothesized that the manifestation of problems may relate to the psychological processes underlying attachment classifications such as expressing and regulating negative emotions. Problems such as risk-taking behaviours, disobedience, social withdrawal or angry outbursts might all be manifestations of the same difficulty in managing negative emotion.

In relation to social functioning, Kobak & Sceery (1988) found that contemporaneous insecure attachment was associated with hostility and lack of social skills in college students. Zimmermann, Scheuerer-Englisch & Grossman (1996) found that security was associated with overall friendship quality in 16 year olds and Allen et al. (1998) found that, in academically at-risk adolescents, attachment security was a significant mediator of the adolescent's social acceptance by peers. Moore (1997) found that secure adolescents were more likely to become sexually active later, had fewer sexual partners and were more likely to use contraception suggesting that securely attached teenagers may be better at forming meaningful romantic relationships in adolescence and that they are also able to negotiate complex social situations such as peer pressure and use of contraception more successfully than their insecurely attached peers.

While the direction of causality may be unclear, the existing research is quite consistent in demonstrating that concurrent attachment style is significantly associated with a number of indices of adaptive and maladaptive psychological functioning in adolescence. There are fewer studies of this design with middle childhood populations. Overall, existing research indicates that it would be useful to complement studies of attachment in infancy and adolescence and psychological functioning with further study of this relationship in the middle childhood stage.

Concurrent attachment: Conclusions.

There is a large and compelling body of evidence that supports the influence of attachment style across the lifespan whether attachment is measured in infancy or at the same time as other indices of psychological and social functioning in adolescents and adults. There is a developing body of evidence supporting this association in childhood. Concurrent attachment research in childhood and adolescence has been complicated by changing attachment behaviour patterns across the developmental period, the lack of appropriate means of measurement as well as the difficulty in determining direction of causality. However, given that attachment has been found to be unstable over time, especially in vulnerable populations, concurrent attachment becomes potentially a more interesting and important research prospect.

1.2.2.1.5 Attachment and disfigurement.

Two key relationships between attachment and disfigurement have formed the basis of much of the research in this area. The majority of studies on attachment and disfigurement have tested the hypothesis that infant disfigurement can act as a risk factor for attachment insecurity. Fewer studies have investigated the role of attachment as a variable that may influence current and later psychological

functioning in young people with disfigurement, although there has been increasing research on this second relationship model more recently.

The next section describes the key research on disfigurement as a risk factor for attachment insecurity, followed by a section that describes the most recent studies on attachment security as a contributing factor to psychological adaptation in people with disfigurement.

Disfigurement as a risk factor for attachment insecurity.

The interaction between appearance and attachment relates to the precursors of the concept of attachment in Konrad Lorenz's ethological studies. These studies indicated that the physical appearance of infants, indeed, of all mammalian species, was associated with the motivation of the caregiver to provide care and proximity, thereby increasing or decreasing the infant's chances of survival into adulthood (Lorenz, 1970).

Early Studies.

A substantial body of small-scale studies from a few decades ago seemed to present a picture of a potentially disruptive and negative effect of disfigurement in newborns on the parent-infant interaction (e.g., De Wolff & Van Ijzendoorn, 1997). Langlois & Sawin (1981) reported that infants perceived as less attractive were held less close to their caregiver and given less contact than more attractive infants from just two days of age. These and other authors hypothesised that if the disfigurement was facial, and particularly if it affected the mouth and eyes, infants may not be able to display emotions effectively, in an easily recognizable way, resulting in difficulties for the caregiver in responding in a sensitive and attuned manner (e.g., Langlois, Ritter, Casey, & Sawin, 1995; Pruzinsky, 1992). The authors hypothesized that this might set off a vicious cycle of the infant initiating fewer emotional expressions

because of poor parental response and the caregiver consequently responding less because of fewer emotional overtures from the baby, ultimately resulting in decreasing opportunities for mutually satisfying and reciprocal interactions. In addition, studies reported that mothers may experience shock and bereavement at the appearance of their disfigured child, which may result in the mother's withdrawal from touching or gazing at their child or being able to experience positive emotions at the sight and presence of their baby which might, in turn, interfere with her ability to respond in a sensitive and attuned way (Walters, 1997) Several early studies of mother-infant interactions in samples of infants with cleft lip and palate pointed to the potentially disruptive effect of the disfigurement on the mother-infant relationship, reporting impairments in both maternal and infant responsiveness (e.g., Barden, Ford, Jensen, Rogers-Salyer, & Salyer, 1989; Field & Vega-Lahr, 1984; Speltz, Goodell, Endriga, & Clarren, 1994; Wasserman & Allen, 1985).

One limitation of these early studies was that they tended to focus on specific behavioural and observable aspects of the mother-infant relationship rather than directly measuring infant attachment itself. The link between observed behaviour and risk of attachment insecurity was extrapolated hypothetically. Although the caregiver's sensitivity and responsiveness to her infant have been shown empirically to be of central importance in determining attachment security, these are not the only factors that contribute to infant attachment security (De Wolff and van Ijzendoorn, 1997). Furthermore, the studies were based on brief periods of observation. As such, maternal behaviour may have altered over time, for example, as the mother adjusted to her baby's diagnosis and appearance.

Murray, Hentges, Hill, Karpf, Mistry, Kreutz, Woodall, Moss & Goodacre (2008) pointed out a number of limitations of the early studies on infants with CLP. Firstly, most early studies were conducted before surgery to repair the cleft had been

carried out (usually done within the first year of life). Babies born with cleft lip and/or palate tend to have multiple concomitant difficulties such as feeding and breathing difficulties and risk of upper respiratory infections. The surgical repair scheduled so early in the infant's life also meant that the infant's early life was dominated by healthcare demands. Furthermore, once the repair had been conducted most, if not all, of the earlier difficulties, as well as the disfiguring effect of an oro-facial cleft, tended to resolve. The early studies did not take the early medical disruptions into consideration. Nor did they follow-up these mother-infant pairs after their repair procedures when their lives became more stable to see if the observed disruptions to the mother-infant interaction persisted or if, indeed, the early disruptions resulted in attachment insecurity. Secondly, raters had not been blind to cleft status, leading to possible biases in ratings. Thirdly, the focus on cleft lip and palate with its very particular medical presentation and course may not be generalisable to other types of congenital disfigurement. This is a limitation that accounts for much of the research on congenital disfigurement; as the fourth most common birth impairment in the U.S accounting for one in 700 live births (Meara & Mulliken, 2011) cleft lip and palate is much more thoroughly researched than other causes of disfigurement.

Later studies.

More recent research has employed more sophisticated methodology to explore the impact of congenital disfigurement on attachment security. Again, these studies are limited by their almost unique focus on the cleft lip and palate population.

In the first large-scale, longitudinal study on attachment security and congenitally disfigured infants, Speltz, Endriga, Fisher & Mason (1997b) began with a cohort of 24 infants with cleft lip and palate, 27 infants with cleft palate only and 64 non-cleft control group infants. The research group planned to measure attachment security over time as well as to identify characteristics in the infant, maternal and social

family domains that predicted attachment security in this population. Attachment was measured at one-year of age using the Strange Situation. In addition, several questionnaire measures were completed, a semi-structured interview was conducted with parents and video recordings of play and feeding interactions were also made.

The results showed highly similar distributions of attachment status across all three groups at one year of age, whether attachment was classified dichotomously (secure and insecure) or into four categories (secure, avoidant, ambivalent, and disorganised). In all three groups, approximately 70% of infants were found to be securely attached. No significant differences were found between the three groups with regard to the distribution of attachment categories, although perhaps surprisingly, infants with cleft lip and palate had the highest level of attachment security among the three groups (80%) and the cleft palate only group had the lowest level of attachment security. The authors concluded that the occurrence of a cleft disorder alone did not appear to significantly increase the risk of attachment insecurity.

Maris, Endriga, Speltz, Jones & Deklyen (2000) reported the results of the follow up to the above study in which the authors investigated the changes in attachment over time. This was thought to be particularly important in the cleft lip and palate population, given that this group experienced several stressors in the first two years of life that could impact on attachment status such as surgical repairs, frequent healthcare appointments and cleft-related medical complications (e.g., with feeding and hearing) and because the surgical repair tended to resolve most of these difficulties quite effectively.

The researchers managed to engage a large proportion of the original cleft sample in the follow-up study losing only five of the original 51 mother-infant dyads. At two years, there were still no significant differences in attachment classifications between the three groups. The control group had the most stable classifications between the first study and the second study, significantly more so than in the cleft palate only group which showed a significant increase in attachment security. The authors attributed this change in attachment security to the typical stressors associated with infants with cleft disorders. The authors suggested that the fact that infants with cleft lip and palate showed (non-significant) higher levels of secure attachment at one year than those with cleft palate only might be because the visibility of the infant's cleft lip might activate more social support mechanisms for parents (compared to the invisible cleft palate) and social support is known to be associated with the development of secure attachment in normally developing infants. The mothers of the infants with cleft lip and palate may receive both more social support and more medical attention around the surgical repair of the lip, which may serve as a "buffer" against the negative effects of the condition on the interaction between the mother and infant.

Whilst this study was helpful in expanding the results of the original Speltz group study, and assessed attachment longitudinally rather than at a single time point, it had a number of limitations. Relatively small within sample sizes meant that findings need to be interpreted cautiously. The reported differences between the cleft palate, and cleft lip and palate groups were not statistically significant. In addition, different coders rated the data at one and two years. Therefore differences in the security rates might be an artefact of different standards of coding, rather than real changes in attachment. Finally, although this study was longitudinal, the timespan across which the participants were followed up was relatively short. Unfortunately, later follow-up data on this cohort have not been published. It would have been useful to

have re-assessed attachment and psychological functioning in childhood to determine the long-term patterns and relationships between these variables.

In the largest UK-based study to date, Murray et al. (2008) also took a longitudinal approach to look specifically at the effect of cleft lip and palate, the timing of lip repair and early mother-infant interactions on cognitive development, behaviour problems and attachment at 18 months of age. Ninety-four infants from four different specialist centres across the UK with cleft lip (with and without cleft palate) and 96 comparison infants were included in the study. The study found that there were no significant differences in attachment security at 18 months of age between infants with a cleft condition and control infants, regardless of the timing of lip repair. At the two-month follow-up assessment of mother-infant interactions, researchers found that the late repair group exhibited the most interaction difficulties. The early repair group was comparable to the control group at the two-month follow-up. However, these differences in interaction difficulties were not found at six or twelve month follow-ups.

The authors provided a brief discussion of why these interaction difficulties are found in the late repair group, suggesting that having a disfigured infant may have a negative impact on maternal mental health, which could affect interaction with the infant. No evidence was found for this hypothesis in this study, however, when maternal depression was assessed. The authors drew on Field and Vega-Lahr's (1984) hypothesis that the cleft disfigurement both made the infant less appealing to look at, and hindered the facial expression of emotions.

While this study is limited in its very short follow-up period (the infants were still only 18 months at follow-up) it is helpful in that it found support for the early studies that reported that disfigurement in infancy might have a negative impact on the mother-

infant interactions. The findings also suggested, consistent with the Speltz study, that once the cleft condition was resolved via surgery, mother-infant interactions appeared normal and there was no discernible difference between clinical and control groups within a relatively short period of time post-repair. These findings appear to be closely associated with the idiosyncratic course and medical treatment of CLP. The question that remains, therefore, is what happens to mother-infant interactions and attachment security if the disfigurement is not largely resolved in infancy as is the case in many other disfiguring medical conditions including dermatological disfigurement. Studies on the impact of long-term disfigurement on attachment over the childhood and adolescent years are still lacking.

A study by Clements and Barnett (2002) is the only study to have compared children with disfiguring conditions with children with another kind of congenital abnormality on a measure of attachment. They compared the relative rates of attachment security in children with congenital neurological conditions and children with congenital disfigurements in a sample of seventy-two toddlers and their mothers. Approximately half of the sample comprised children with neurological abnormalities (e.g. epilepsy, cerebral palsy) and the rest had non-neurological birth abnormalities, including disfigurements (predominantly cleft conditions, but also a minority with other types of craniofacial anomalies). The child's attachment status was assessed using the Strange Situation and the Attachment Q-sort (Waters & Deane, 1985) depending on their age at the time of assessment. Parenting quality was assessed from a videotaped parent-child play session. The severity of the child's disfigurement was also recorded.

The study reported that diagnostic group was not related to attachment classification when all insecure attachment classifications were analysed separately, but a significant relationship emerged when a two-way secure/insecure attachment

classification was used. Children with non-neurological congenital anomalies (including disfigurements) were significantly more likely to have secure attachments than children with neurological disorders. No comparisons to a normative population were made. Attachment security in both groups was associated with a significantly higher quality of parenting. Parenting quality was found to be a weak but significant predictor of attachment status, when the child's condition was controlled for. Parenting quality was most strongly predicted by the "appearance impact" (i.e. severity) of the condition. More apparent physical anomalies appeared to elicit higher quality parenting. The authors suggested that a degree of vulnerability in the child might actually enhance parenting sensitivity whereas a much higher level of need in the child, as in the case of children with neurological abnormalities, might be problematic to parental sensitivity.

Two studies have researched adults with disfiguring dermatological conditions, attachment and psychological functioning (Picardi et al., 2005; Rabung, Ubbelohde, Kiefer, & Schauenburg, 2004). The findings of these adult studies contradict those of the studies based on infants with oro-facial anomalies in finding an increased rate of attachment insecurity in the dermatologically disfigured group. These papers are described in detail in the next section.

Conclusion

In summary, the studies considered thus far have not found higher rates of insecure attachment in infants with cleft-related disfigurements compared to non-clinical comparisons. However, as highlighted throughout, there are a number of methodological limitations in these studies, making these results tentative, and difficult to generalise to the wider population of disfigured children. Because of their exclusive focus on the CLP population, the existing studies do not inform us about children who suffer from congenital disfigurements that are not largely resolved in

the first year of life and that have a different constellation of symptoms and a different disease course. Furthermore, these studies have only measured attachment in very young children, the oldest cohort being no more than three years of age.

Attachment as a risk factor for psychological dysfunction in people with disfigurement.

The theory that attachment security may act as a protective factor or insecurity as a risk factor in the psychosocial functioning of young people with disfigurements is based on the hypothesis that having a disfigured appearance, itself, constitutes a psychosocial risk due to the increased likelihood of social stress in the form of:

1) Implicit social rejection (such as social exclusion, stigmatization, devaluation)
2) Explicit social rejection (such as bullying, victimization)
3) Ambiguous but intrusive social situations (such as being stared at or questioned by strangers).

In combination with the comprehensive empirical support for attachment being significantly associated with psychological functioning in the general population, it was hypothesized that a combination of attachment-based processes such as internal working models about the intrinsic value of oneself and positive or negative expectations of others, emotion regulation, reflective capacity and learnt social behaviours might protect a disfigured young person from the impact of negative social experiences or, conversely, exacerbate the effect of negative social experiences.

While no studies have yet been published on the role of attachment in the psychological development of children with disfiguring dermatological conditions, a

few studies have been conducted on adults with disfiguring dermatological conditions. These studies tested if the sample with a disfiguring dermatological condition was more likely to report insecure attachment as well as if attachment security was associated with psychological function.

A German study conducted by Rabung, Ubbelohde, Kiefer & Schauenburg (2004), investigated attachment style in 124 adults with atopic dermatitis using the German version of the Relationships Styles Questionnaire (RSQ) (Steffanowski, Oppl, Meyerberg et al., 2001). Attachment style was further covaried with disease severity, depression, anxiety, social support and quality of life. The authors reported that there was a significantly increased rate of attachment insecurity (54%) in their clinical group compared to German norms (39%). The paper further reported that participants who reported disease onset in the first year of life were more likely to report insecure attachment style than those who had acquired their disease in adulthood. However, this difference failed to reach statistical significance. Disease severity was equivalent across attachment categories. However, attachment security was significantly associated with more social support and better quality of life as well as significantly lower levels of depression and anxiety. Unexpectedly, a stronger association was found between disease severity, social support and life quality in the securely attached group in a multiple regression analysis. The authors hypothesized that the insecurely attached group might have been more vulnerable to stressors other than their skin disease therefore reducing the association between skin disease and psychosocial functioning.

There are empirical limitations to this study that should be considered. First, as a cross-sectional study, it is impossible to determine causal relationships. Second, while the RSQ has been found to demonstrate good reliability and validity, because it is a self-reported measure, there is a risk of shared method variance accounting

for associations between measures. Third, it is impossible to determine the stability of attachment over time: if attachment style was affected by the experience of chronic illness either in infancy or over the lifespan. The authors allude to possible usefulness of further investigation in reporting the non-significant difference that participants who acquired their dermatitis in the first year of life were more likely to report insecure attachment in adulthood than participants who acquired their dermatological condition later in life. That is, that having a dermatological condition in the key phase associated with development of attachment style; the first two years of life, may have increased the likelihood of attachment insecurity.

An Italian research team interested in exploring the common belief that stress can trigger a number of dermatological conditions investigated adults with Alopecia Areata (Picardi et al., 2003a), Vitiligo (Picardi, Pasquini, Cattaruzza, Gaetano, Melchi, Baliva, Camaioni, Tiago, Abeni, & Biondi, 2003b) and Psoriasis (Picardi et al., 2005). The researchers collected data on life stresses experienced in the year preceding disease onset or relapse. They also collected data on what they considered to be potential stress moderating variables: attachment style (Experience of Close Relationships Scale (ECRS); (Brennan, Clark, & Shaver, 1998) emotional style (Toronto Alexithymia Scale (TAS); (Bagby, Parker, & Taylor, 1994) and social support (Multidimensional Scale of Perceived Social Support (MSPSS);(Zimet, Dahlem, Zimet, & Farley, 1988).The studies each reported very similar findings across dermatological conditions. They consistently found that participants with dermatological conditions did not experience more stressful life events than the control group prior to a flare up of their condition. However, in all three studies, the experimental group was found to report significantly higher rates of avoidance in their attachment relationships, higher rates of emotional difficulties and lower social support. These findings led the authors to conclude that the onset of dermatological conditions was not so much related to the quantity of life stress

experienced by these individuals but the way in which they responded to stressful events which was, to some extent, related to their attachment style. It should be noted that each clinical group was relatively small in size and that the research design was cross-sectional. Replication of these studies with a larger population using a longitudinal design would be valuable.

Schmidt, Nachtigall, Wuethrich-Martone & Strauss (2002) explored attachment and illness coping style in patients with a range of chronic medical conditions, one of which was the disfiguring dermatological condition, Alopecia Areata, the main feature of which is idiopathic hair loss. The authors reported a moderate association between adult attachment and illness coping style. The insecurely attached participants exhibited what the authors referred to as “inflexible coping”, leading the authors to conclude that secure attachment was likely to be an important internal resource for adaptation to a disfiguring illness.

Conclusion

While these studies demonstrate strong evidence for attachment insecurity to be associated with disfiguring dermatological conditions in adults and with increased psychological difficulties in this population, they do not help us to understand causal direction because of their cross-sectional design. Furthermore, these findings based on adult participants cannot be directly extrapolated to child populations as it is still unclear whether psychological processes in childhood are similar to psychological processes in adulthood. Nor do studies with adults help us to understand the pathway relating attachment security and dermatological disease in adulthood. Further research on child populations would help to understand the association between attachment and disfiguring dermatological conditions in this earlier developmental phase as well as, potentially, shedding light on the relationship between attachment and disfiguring dermatological disease over time.

Attachment and disfigurement: Conclusions.

In considering the broader literature, there is justification for proposing an association between attachment and disfigurement. First, there is theoretical support for the hypothesis that having a very unwell or unattractive infant could impede a mother's ability to interact closely with their baby thus affecting attachment security (e.g., Lorenz, 1970). There has been little support for disfigurement as a risk factor for attachment insecurity in studies based on infants with CLP (e.g., Speltz et al., 1997; Murray et al., 2008). However, studies on the effect of disfigurement on attachment have been limited by the fact that they have focused solely on children with oro-facial anomalies such as CLP. Furthermore, any follow-up of children with congenital disfigurement has been only after a very brief amount of time and while the affected child is still an infant. No studies have investigated older children with lasting disfigurement. In contrast, studies on adults with disfiguring dermatological conditions have found increased rates of attachment insecurity, an association between attachment and disease manifestation, and an association between attachment security and a range of psychological indices including adjustment to illness, quality of life and social support (e.g., Picardi et al., 2005; Rabung et al, 2004). These findings are yet to be replicated with a child sample.

Second, there is also an argument for attachment insecurity to increase the likelihood of disease (e.g., Schmidt et al, 2002; Picardi et al, 2005) and psychological dysfunction (e.g., Allen, Moore, Kuperminc & Bell (1998) in adulthood. Attachment has been shown to be a significant risk or resilience factor in the general population, especially in the context of increased psychosocial risk. Since childhood chronic illness or disfigurement can be construed as a risk factor for psychological difficulties (Hamilton, 2000) one can hypothesise that attachment could also be a

significant factor in the development of a child with a chronic, disfiguring condition. While the small number of studies on adult populations have found promising results, there has been no published research, as yet, that has investigated this causal pathway in children and adolescents with disfiguring dermatological conditions.

The empirical support for attachment style to be a significant factor in the psychological functioning of people with disfiguring dermatological conditions is still in its infancy. Questions remain, particularly with regard to children and adolescents with congenital disfiguring dermatological conditions. Empirical support is, overall, still weakened by limited sample characteristics, small sample sizes and a small number of studies.

1.2.2.1.6 Attachment and disfigurement: The current study.

The current study aimed to fill some of the knowledge gaps in the existing literature on attachment style and disfigurement. In relation to attachment there were two aims.

The first aim was to determine if children and adolescents with congenital dermatological disfigurement are more likely to be insecurely attached than children in the general population. That is, if congenital dermatological disfigurement might act as a risk factor for attachment insecurity. Congenital rather than acquired dermatological disfigurement was of particular interest in relation to this first aim as participants would have had their dermatological condition during the key attachment formation period of the first two years of life. Based on the studies of adults with disfiguring dermatological conditions, it was hypothesized that children with congenital dermatological disfigurement would show a higher rate of attachment insecurity.

The second aim of the current study was to determine if concurrent attachment style is associated with socio-emotional functioning in later childhood and adolescence. While there is a sizeable body of literature that supports the importance of infant attachment in relation to later psychological and social variables, the current evidence suggests that it would be unsafe to assume attachment continuity from infancy through to childhood, adolescence and adulthood (Weinfield et al., 2000) especially in individuals affected by a chronic medical condition. As such, studies based on infant attachment may not be relevant to studies on concurrent attachment. There is very limited research on the association between concurrently measured attachment and psychological functioning in middle childhood. However, studies on non-disfigured adolescents and on a range of adult populations, including adults with dermatological disfigurement suggest a significant association between concurrent insecure attachment and poorer psychological functioning on a range of indices. As such, in the current study, it was hypothesized that concurrent attachment style would be significantly associated with indices of socio-emotional functioning in children and adolescents with congenital dermatological disfigurement.

1.2.2.2 Shame

Shame is a construct that has been the focus of a recent renewed interest in empirical psychology in the context, primarily, of the cognitive behavioural therapies referred to as Third-Wave CBT, namely compassion-focused therapy (e.g., Gilbert, 2009). Shame can be defined, briefly, as the emotional, cognitive and behavioural response to the experience of actual or perceived social rejection or devaluation (Gilbert, 1998b). It is described as the multi-modal response based on the sense that one is seen as a fundamentally bad person rather than someone who has

committed a bad act (Lewis, 1971). High or persistent experiences of shame have been associated with psychopathology in adults (eg. Gilbert, 2000).

In the current study, shame was considered to be a possible contributing factor in determining socio-emotional functioning in young people with congenital dermatological disfigurement. The reason for including shame in the explanatory model was that disfigurement was seen to increase the risk of social rejection and devaluation due to the role of appearance in social interactions. As such, it was hypothesized that disfigured young people would be at increased risk of experiencing shame. Because shame has been associated with psychopathology in adults, it was hypothesized that increased shame due to increased social rejection might be one pathway via which disfigurement might be linked to increased psychopathology. However, shame has yet to be comprehensively investigated with child and adolescent populations.

The first part of this section will provide:

1) A brief descriptive background on the construct of shame.
2) Conceptual and empirical support for a relationship between shame and maladaptive psychological functioning.
3) Examination of the evidence for two hypothesized relationships between shame and disfigurement i) that congenital disfigurement will increase the likelihood of dispositional shame ii) that shame will be positively associated with psychological maladjustment in young people with congenital dermatological disfigurement.

1.2.2.2.1 The Phenomenology of Shame

The concept of shame has been explored in the western psychological literature for several decades (e.g., Goffman, 1955) but is significantly pre-dated by its

description as being integral to developing adaptive social functioning in collectivist societies for millennia. Confucius, whose writings have been dated to approximately 500 years before the common era, was reported as saying that shame was an emotion that drove one's focus inwards for self-examination and that it motivated the person towards change (Li, Wang, & Fischer, 2004). In Japan and China, shame and self-criticism are still used as predominant socializing forces in child rearing (e.g., Fung, 1999; Lewis, 1995). Indeed, shame in the socio-anthropological literature, in a range of social groupings, collectivist or otherwise, is largely viewed as an adaptive emotion where it is broadly conceptualized as an internal warning signal that serves to facilitate, rather than impede, socially and personally adaptive behaviour. As such, shame has been described as playing an essential part in promoting inclusive fitness through the regulation of behaviours that are crucial in the areas of group identity, social bonding and competitive mating success (Greenwald & Harder, 1998).

In the modern psychological literature, shame has gained renewed empirical and clinical interest, but as a maladaptive trait. Research has focused on the association between shame and a range of psychopathology such as depression (Gilbert, 2000; Tangney, Burggraf, & Wagner, 1995) anger and violence (Dutton, van Ginkel & Starzomski, 1995), anxiety (Gilbert, 2000) and post-traumatic stress disorder (Lee, Scragg, & Turner, 2001).

The term shame has been used to describe phenomena in many different forms including internal self-experience, relational episodes and even cultural practices relating to honour and prestige. Gilbert (1998a) summarized what he considered to be the key facets of the shame phenomenon:

1) A social or external cognitive component. Shame affects are said to be elicited in social contexts and to be associated with the perception that others see the self as inferior, bad, inadequate and flawed. That is, that others would look down, or are looking down, on oneself with a denigrating or contemptuous view

2) An internal self-evaluative component. For many theorists shame involves a global negative self-evaluation as bad, inadequate or flawed. Shame is thus commonly associated with negative automatic thoughts about the self. Indeed, many self-critical and self-attacking thoughts are in essence self-devaluations and therefore internally shaming thoughts. This aspect of the shame experience has received the most empirical attention (Gilbert & Miles, 2000)

3) An emotional component. Several theorists support the idea originally mooted by Tomkins (1981) that shame is an emotion that binds with other primary emotions such as fear, anger or disgust, giving different facets to how shame is experienced. However, many people can describe a unique emotional experience characterized by a sense of inner deflation or dejection (Gilbert & Mcguire, 1998). Alternately, some theorists have conceptualized shame as the interruption and sudden absence of positive affect (Nathanson, 1992).

4) A behavioural component. Shame is often associated with specific defensive behaviours such as a strong urge to not be seen, to avoid exposure, to conceal or to run away (e.g., Tangney et al., 1995). Eye gaze is commonly averted and the individual may feel behaviourally inhibited. These responses have been linked to a rapid onset of submissive defensive behaviours seen in a range of higher order mammals (e.g., Darwin, 1872); Keltner & Harker, 1998).

5) A physiological component. Shame is conceptually related to a stress response although the exact nature of it is unclear. In some cases it may involve heightened parasympathetic activity (e.g., Schore, 1998). Although our knowledge of the neurophysiology of primary emotions, such as anger, anxiety and disgust is increasing, less is known about the neurophysiology of shame.

Therefore, shame can be conceptualised as a complex set of feelings, cognitions and actions; tendencies whose exact manifestation can vary between individuals and across different social contexts.

External and Internal Shame

Gilbert believes that shame can be grouped into two categories; external shame and internal shame (Gilbert, 1998a) Gilbert described external shame as the response based on the perception that one exists in the mind of others in a negative or devalued way. Gilbert viewed this type of shame as more akin to the Tangney concept of the self-conscious emotions in that it is based on the human ability to understand what might be in the minds of others sometimes referred to as “theory of mind” (Gilbert, 2002). Gilbert explained that external shame can occur when the individual experiences actual social rejection, but that external shame can also be elicited by the fear or anticipation of social rejection. Gilbert explained that several different emotions could be elicited by this cognitive-affective response such as anxiety or sadness as well as shame and guilt. Gilbert believes that typical defensive behavioural reactions would include concealment, avoidance, and social withdrawal. External shame represents beliefs that others judge the self negatively, which may result in threatened or actual loss of value to the group or social rejection (Gilbert, 2002). Here the focus is on the outside world and how one is perceived by others (Gilbert, 1998a).

In contrast, Gilbert coined the term internal shame to describe the process of devaluation by oneself rather than by others; when the individual believes and feels that they are fundamentally bad, weak or undesirable. Gilbert explained that internal shame was clearly related to other cognitive concepts such as negative self-schema (Beck, 1979) and low self-esteem (Fennell, 1997) and appeared to share similar

underlying cognitive processes such as appraisal (Smith & Lazarus, 1993). To date, it is still unclear, however, how internal shame relates to these other cognitive processes; if, for example, both shame-proneness and negative self-concept are caused by the same early developmental experiences such as misattunement in the attachment relationship or early childhood devaluation via child maltreatment or if one causes the other (Gilbert, 1998a). Gilbert (1998a) added that internal shame is experienced when a failure to meet social standards is attributed to the self, specifically when it involves global negative self-evaluation and beliefs about the self as intrinsically bad or flawed.

The concept of internalised shame is not unique to Gilbert. Kaufman (1989) described the process by which stigma and shame can be internalised in the form of scenes and scripts and memories of feelings and experiences of being shamed. Kaufman hypothesized that individuals were most vulnerable to internalizing shame when an individual's fundamental need for love, affiliation and belonging and status is thwarted by the loved ones that they most depend upon to affirm their sense of value and attractiveness, such as parents.

Although external and internal shame have often been found to be highly correlated (Goss, Gilbert, & Allen, 1994b), Gilbert proposes that the two are distinct, but related constructs (Gilbert, 2002). Repeated experiences of negative and devaluing messages from others, associated with external shame, might contribute to the development of internal shame, and a state of internal shame might increase the likelihood of the individual interpreting the behaviours of others as rejecting therefore externally shaming. However, internal and external shame have been found to be elicited and to act independently of one another (Camp, Finlay, & Lyons, 2002).

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1.2.2.2.2 Development of Shame

Shame is universally accepted as the emotional reaction relating to devaluation of the self in the social context. In order to understand the role that shame might play in positive or negative psychological adjustment in congenitally disfigured young people it is helpful to understand the means by which shame develops.

The primary attachment relationship has been viewed as the key factor in the development of adaptive and maladaptive shame responses in young people. Experts in the field theorise that attachment may drive the shame response in two ways. The first pathway is that of emotional regulation. Proponents of this pathway argue that shame, the emotional reaction to normal, periodic, maternal misattunement, can be an adaptive learning opportunity, in a secure attachment relationship, whereby periods of misattunement are brief and the infant is supported by the attachment figure to learn how to tolerate and then overcome those unpleasant feelings, returning back to a state of positive affect rapidly. In an insecure attachment relationship, it is argued that infants experience chronically misattuned interactions from their primary attachment figure, or where shame is experienced, the infant is not supported in returning to a positive state. Therefore, the shame experience is experienced as even more unpleasant and prolonged than in the securely attached infant (e.g., Schore, 1998).

Theorists also argue the role of attachment in developing Internal Working Models (IWMs) relating to the self and others based on early attachment experiences. Secure attachment relationships are thought to contribute to the development of IWMs that view the self as loved and valued and of others as kind and responsive, while insecure attachment experiences are thought to lead the infant to a view of self that is rejected and unworthy and others as punitive and distant. These negative

cognitive structures would elicit a shame response (e.g., Yelsma, Brown & Elison, 2002) and ultimately psychopathology in later life (e.g., Hankin & Abela, 2005).

Later experiences in the broader social contexts of the family and community have also been hypothesized to have an influence on the formation of an individual's shame response. Several studies has found an association between criticizing and rejecting messages from parents with shame-proneness in adulthood (Gilbert, Allan, & Goss, 1996a; Mills, Nazar, & Farrell, 2002; Alessandri & Lewis, 1996b; Kelley, Brownell & Campbell, 2000). A large body of empirical work also supports the role of implicit and/or explicit social stigma (Lewis, 1996) on shame (e.g., Thompson et al., 1995; Cattarin & Thompson, 1994)

Due to the theorized roles of early attachment relationships and rejecting or devaluing messages in the social context on shame, this construct was considered as potentially important in understanding the impact of social rejection on young people with congenital dermatological disfigurement. However, these compelling theoretical arguments are yet to be strongly supported in the empirical literature.

Summary of Conceptual Literature on Shame

The concept of shame is a very old one, which has been described in a number of cultures and across millennia (e.g. Confucius in Li et al., 2004). Shame has re-emerged in the last century in the mainstream social sciences, including psychological research, more recently becoming the focus of new developments in cognitive behavioural therapies in the form of compassion-focussed cognitive therapy (Gilbert & Irons, 2005). However, the definition of shame, the way in which it develops and how it shifts from a normative to a pathological process is still difficult to operationalise.

Shame is seen by all theorists in the field as a typically normative and adaptive process that facilitates social inclusion. However, in certain people, shame can reach pathological levels of intensity or frequency, called dispositional shame, in the same way that fear can become pathological in the form of anxiety. Shame in this pathological form has been associated with a range of psychological problems and difficulties in engaging in interventions for those problems (e.g., Gilbert & Irons, 2005).

There is a developing body of literature that attempts to operationalise the phenomenon of shame. However, a few key empirical issues that are yet to be resolved hamper the progress of this field of research substantially. First, difficulties in measuring shame, distinct from other psychological phenomena such as other affective states and cognitive structures means that it is currently very difficult to be sure that a study that purports to be measuring shame is measuring shame and not something else. While many studies show strong associations between shame and a range of psychological difficulties, it is difficult to determine unequivocally if these studies are not, in fact, measuring the same or closely related psychological phenomenon. Second, much of the research is, at best correlational. As such, it is impossible to distinguish psychological phenomena chronologically and how the variables relate aetiologically. Much research associating shame or hypothesized shame-inducing experiences in early life is conducted retrospectively. The participants often include individuals with a known history of significant psychopathology. As such, it is difficult to know if their recollections of their own development are objectively accurate, filtered by distorted psychological processes or biased by current life situations and perceived advantage or disadvantage in reporting. Finally, much of the research in early childhood is based on observing behavioural indices assumed to be associated with shame such as postural or facial expressions and expressions of emotional distress. It has, so far, been impossible to

be certain that shame is, in fact, what that individual is experiencing. Furthermore, very few studies have been conducted on shame in middle childhood and adolescence, particularly in relation to psychopathology.

In summary, there appears to be strong prima facie support for the phenomenon of shame and the way in which it has been described. However, the empirical research is yet to provide an unequivocal and clearly differentiated picture of shame.

Despite the conceptual and empirical inconsistencies in the shame literature, because of the central role of shame in social rejection, a core premise underpinning the current study, shame was considered to be important to the current study.

For the purpose of the current study, shame was defined as a multi-modal response to the experience or perception of social rejection or devaluation (Gilbert, 1998). Shame was conceptualized as a primary emotion that manifests itself in emotional, behavioural and cognitive phenomena (Gilbert, 1998). Shame was described as encompassing two related but distinct types of shame: external shame defined by Gilbert (2000) as the reaction to the sense that one is seen by others to be of lower status or value and internal shame as that sense of feeling devalued by other adopted or internalised by the individual so that they also view themselves in a devaluing and rejected way.

1.2.2.2.3 Shame and Psychopathology

There have been a number of pathways proposed to explain the relationship between shame and psychological adjustment. The most robustly researched models have tended to be those with a cognitive basis.

Cognitive theorists (e.g., Reimer, 1996) have proposed that the internal, global and stable attributions that are associated with shame such as “I am not as good as other people” predispose the individual towards developing psychological difficulties such as depression (Seligman, Abramson, Semmel, & Baeyer, 1979). Brewin (1996) provided support for the association between cognitive structures and the experience of shame. Brewin proposed the cognitive vulnerability model in which negative self-schema elicited shame feelings which, in turn, reinforced the negative self-schema. Brewin admitted that the exact pathways via which this process occurred were still unclear but that there was already clear support for the role of each negative self-schema and dispositional shame in a number of psychological outcomes. For example, much has been written about the overlaps between shame and social anxiety; fear of being negatively evaluated by others, perception of self to be socially unattractive to others and the typical defensive behaviour of avoidance and social withdrawal (e.g., Gilbert, 2000).

Gilbert & Procter (2006) described a cognitive-affective model, which linked the pathogenic qualities of shame to two key processes. The first quality was the degree of self-directed hostility, contempt and self-loathing associated with self- criticism (Gilbert, 2000; Whelton & Greenberg, 2005; Zuroff et al., 2005). Second was the relative inability to generate feelings of self-directed warmth, soothing, reassurance and self-liking (Gilbert, 2000; Gilbert, Clarke, Kempel, Miles, & Irons, 2004; Linehan, 1993; Neff, 2003a; Whelton & Greenberg, 2005). The authors described shame as having a “stickiness” that could easily result in a ruminative, self-critical style, which would increase vulnerability to a range of psychological difficulties. Gilbert & Miles (2000) described shame as being transdiagnostic, permeating many disorders, increasing vulnerability to psychopathology, symptom expression and risk of relapse.

Scheff (2012) placed shame at the interface between the self and the social context. Scheff explained that shame acts as a feedback loop whereby the individual who finds themselves experiencing a stigma such as mental illness or disfigured appearance experiences shame as a result of their expectation of how others will respond to knowledge of their stigma. This then leads to a downward spiral of negative emotion leading to mental illness, which can also include feelings of shame about feeling shame.

Again, while promising empirical work has begun to deconstruct and describe the shame pathways reliably, there is still little concrete evidence for the hypothesized processes that underpin shame, many of the existing studies investigating associations between variables rather than explicit causal relationships.

Shame and Psychopathology in the Non-disfigured Population: Empirical Evidence

There is a large body of research that demonstrates significant associations between shame and a range of psychological difficulties in the non-disfigured population. Shame with its focus on the fundamentally defective nature of the self is closely associated with various maladaptive response tendencies such as anger and externalizing in middle school-aged children (Tangney, Wagner, Hill-Barlow, Maschall & Gramzow, 1996). Studies with adults and adolescents have revealed moderate to strong links between shame and symptoms of psychopathology such as depression and anxiety (e.g., Tangney, 1990, 1991, 1994)

Shame, Depression and Anxiety

In the adult population, significant correlations have been found between external shame and clinically significant depression in adults. This research also showed that external shame accounted for 33% of the variance in depression scores (Gilbert, McEwan, Bellew, Mills, & Gale, 2009). In one of the very few studies on young

people, over 300 Singaporean adolescents were sampled from a small number of government schools. Internal shame was found to be significantly correlated with psychological functioning (Ang & Khoo, 2004). Participants in the co- morbid anxiety, depression and aggression group scored significantly higher on internal shame than those in the single disorder (anxiety, depression or aggression) groups, who in turn scored significantly higher than those in the control (no diagnosis) group. However, it was not clear to what extent the findings could be generalised since the culture specific experiences of shame have been well argued (Reimer, 1996), and because there were only 10, 11 and 12 participants in the co-morbid, aggression-only and anxiety-only clinical groups, respectively, with a more substantial 65 participants in the depression-only group.

Hughes, Gullone & Watson (2011) ran a large-scale study with 9-15 year olds comparing one group who were reporting depressive symptoms and a control group reporting no depressive symptoms in an attempt to identify emotional functioning processes that may underlie the experience of childhood depression. Shame-proneness was one of the emotional variables that was found to be significantly associated with depressive symptomatology along with poorer emotion regulation. Again, because the data were collected contemporaneously, it was impossible to determine is there was a causal relationship between shame and the other variables.

Studies have shown significant correlations between internal shame and anxiety in adolescents (Ang & Khoo, 2004) and adult inpatients and outpatients (Gilbert, 2009). In a study of 64 adults receiving mental health treatment, external shame was found to be strongly correlated with anxiety $r(64)=0.54$, which was significant at an alpha level of 0.01 (Gilbert et al., 2009). As with depression, the cross sectional methodology used meant that a causal effect could not be determined.

1.2.2.2.4 Shame and Disfigurement

The existing literature suggests two possible ways in which shame and disfigurement might be related. Firstly, shame has been found to be associated with a range of psychopathology in non-disfigured populations. As such, it is possible that shame will be associated with psychopathology in a disfigured population also. If this is the case, then clinical interventions that address shame may be indicated when working with disfigured clients with psychological problems. Secondly, research has suggested that disfigurement may result in high shame in affected individuals because the bio-evolutionary value that humans place on attractive physical appearance will result in experiences of social rejection and feelings of devaluation and stigmatization in disfigured individuals.

The conceptual and empirical evidence for both of these proposed relationships is discussed below.

Disfigurement as a cause of shame

There are many writers who make theoretical and empirical arguments for the importance of shame in individuals with a disfigured appearance (Kent & Thompson, 2002; Thompson & Kent, 2001) in a range of clinical populations including adults with psoriasis (Miles, 2002) and burns (Coughlan & Clarke, 2002). These arguments are based on the empirical findings presented in Section 1.2.1.4, that disfigured appearance elicits a negative response from others. Several authors have discussed disfigured appearance as a stigmatizing attribute that results in social devaluation and rejection that can be explicit or implicit.

As social rejection, or the threat of social rejection, is hypothesised to be one possible pathway to shame (Gilbert & Andrews, 1998), this research implies that

individuals with a disfigured appearance would be more likely to experience external shame. For some, this shame may become internalised (Eaton, 1999).

Disfigurement as a cause of shame in young people: Empirical Evidence

There are few published studies on shame in disfigured children and young people. In an unpublished study, Eaton (1999) compared 16 adolescents with cleft lip and/or palate (CLP) with 16 healthy control participants on shame-proneness, focus of shame and bullying. Using a questionnaire measure developed by the author, the study reported that while the CLP population reported facial appearance as the focus of shame significantly more than their healthy counterparts, there was no significant difference in shame proneness between the two groups. There was also no significant difference in the amount of bullying reported by the two groups. High bullying was associated with higher shame-proneness in both CLP and comparison populations.

The Eaton (1999) study provided support for the argument that bullying, as a form of explicit social rejection, is associated with shame-proneness but refuted the hypothesis that disfigurement would increase shame or bullying. However, these findings should be treated cautiously. Firstly, the sample of this study was very small; 16 adolescents in each group and it is, therefore, possibly not representative of the CLP population. Secondly, the study focused only on young people with CLP which limits generalisability to young people with other forms of congenital disfigurement. Thirdly, because the data were collected simultaneously, it was impossible to determine the causal relationship between shame-proneness and bullying; are shame-prone individuals more likely to attract bullying behaviour or to interpret social interactions as bullying or are individuals who are the victims of bullying more likely to report shame-proneness. Finally, the study potentially suffers from shared method variance in that all measures were self-reported and the views

of other key people around the young participant were not surveyed.

Shame and Psychopathology in Disfigured Young People: Empirical Evidence

Two studies have directly explored the relationship between shame and psychological adjustment in a population of adolescents with disfiguring dermatological conditions. Soon & Harvey (2009) studied shame and psychopathology in 78 adolescents with disfiguring dermatological conditions. The Internalised Shame Scale (ISS: (Cook, 1988) was used to measure internal shame and the Other as Shamer scale (OAS: (Goss, Gilbert, & Allan, 1994a) measured external shame. Psychopathology was measured using the Emotional Symptoms subscale of the Strengths and Difficulties Questionnaire (SDQ: (Goodman, 1997) and the self-esteem subscale of the ISS. Parents of the teenage participants were asked to complete a social support measure and a measure of the importance of appearance as well as completing the parent-report version of the Emotional Symptoms subscale on the SDQ.

The results confirmed the hypothesis that shame was significantly related to the Emotional Symptoms score in this population. The associations were significant for both parent-report and self-report versions of the SDQ. Significant associations were also found between both internal and external shame, social support and self-esteem. Neither internal nor external shame were associated with the importance of appearance measure. As a non-disfigured control group was not employed in this study, it was not possible to draw conclusions about whether the levels of shame reported by the adolescents with disfigurement were different from those in the general population. Also, because the data were collected at the same time it was not possible to determine if there was a causal relationship between shame and emotional symptoms. Because self-reported shame was significantly associated with parent reported psychological outcome, as well as self-reported psychological

outcome, it was considered unlikely that the association found between shame and psychopathology was solely due to shared method variance.

In a follow-up study, Mason, Brown & Soon (2010) compared a group of adolescents with disfiguring dermatological conditions with a school-based non-disfigured control group and a group of adolescents with a non-disfiguring medical condition, Phenylketonuria (PKU). External shame was found to have the strongest relationship with the psychological outcome variables of depression and anxiety with R^2 change values of .16 and .42 respectively. However, contrary to expectations, the healthy control group reported significantly higher levels of internal and external shame compared to the disfigured group and the PKU group. The disfigured group reported significantly lower levels of shame than the other two groups. The non-disfigured groups also reported significantly higher levels of anxiety and depression suggesting that the groups may not have been comparable in terms of overall psychological functioning.

Sampogna, Tabolli & Abeni (2012) conducted a large-scale study (N=786) with adults with psoriasis. Using the Italian version of the Skindex-29 questionnaire (Abeni, Picardi, Pasquini, Melchi, & Chren, 2002), a measure of quality of life in dermatology patients, and the General Health Questionnaire (GHQ; (Goldberg, 1988) the most commonly reported emotional difficulties were shame, anger and worry. Elevated scores on these three emotions were associated with clinical anxiety and depression. As with the Soon & Harvey (2009) study, the study did not include a non-disfigured comparison group and was, therefore, unable to determine if psoriasis sufferers were more likely to experience shame than the healthy population.

A recent study conducted in Germany compared 87 children with facial burns, haemangiomas, port-wine stains or congenital melanocytic naevii (CMN) with a community-based non-disfigured control group on experiences of stigma (Masnari, Schiestl, Rossler, Gutlein, Neuhaus, Weibel, Meuli, & Landolt, 2013). The study also tested a number of psychological and disfigurement variables to identify the factors accounting for most of the variance in reported stigma experience. The authors, using the German language version of the Perceived Stigmatisation Questionnaire (PSQ)(Lawrence et al, 2006) found that the disfigured group reported significantly higher perceived stigmatization scores than the non-disfigured comparison group. The variables identified in the disfigured group that accounted for the most variance in stigmatization were age (the older the child the more stigmatization reported) and the size of the disfigurement, with children whose disfigurement covered more than 25% of their face reporting more stigmatization. However, of the 27% of variance accounted for by these two variables combined, size of disfigurement only accounted for 5%. A key strength of this study was the inclusion of a control group sample, which allowed the authors to conclude that disfigured children were significantly more likely to experience stigmatization than children in the control group. However, the study did not measure shame or any other internal psychological factors. As such, it is impossible to understand exactly how perceived stigma might be related to shame and other psychological variables.

Disfigurement and Shame: Conclusions

Two relationships between disfigurement and shame have been investigated so far. First, that having a disfigured appearance would lead to increased shame. Second, that shame, in individuals with a disfigured appearance, would be associated with greater psychological difficulties.

The conceptual literature provides a strong argument for the notion that having a disfigurement significantly increases the risk of reporting dispositional shame. However, the only studies measuring shame in young people with disfigurement did not find support for increased shame in this population compared to non-disfigured comparison groups of the same age range (Mason, Brown & Soon, 2009; Eaton, 1999) On the other hand, Sampogna et al (2012) found significantly increased shame in their group of adults with disfiguring dermatological disease and Masnari et al (2013) found significantly increased reports of perceived stigmatisation in their group of children with disfiguring skin conditions. Overall, the relationship between shame and disfigurement in young people is still unclear.

The second relationship between shame and disfigurement - that shame will be associated with increased psychological dysfunction in people with disfigurement - has received more empirical support (i.e.,(Mason et al, 2009; Soon & Harvey, 2009; Sampogna et al, 2012; Masnari et al, 2013) finding a strong association between shame and psychological outcome indices in the form of measures of depressive and anxious symptoms and self-esteem. This research is further supported by studies on non-disfigured populations where shame is consistently found to be associated with poorer psychological outcomes (e.g., Tangney, Wagner, & Gramzow, 1992).

In conclusion, there is a strong theory-based rationale for shame being a key psychological factor in people with disfigurement. However, at this time, the empirical support for shame to be higher in people with disfigured appearance is equivocal. The empirical evidence is far more compelling with regard to shame being associated with poorer psychological outcome in both non-disfigured and disfigured populations. There is clearly further empirical work to be done in understanding the relationship between disfigurement and shame.

1.2.2.2.5 Shame and Disfigurement: The Current Study

The current study aimed to develop the existing evidence by replicating some initial studies conducted on shame and disfigurement. First, the current study aimed to test the hypothesis that young people with congenital dermatological disfigurement would report higher levels of internal and external shame than non-disfigured young people. This hypothesis is held because of the strong theoretical support for the hypothesis that people with a disfigurement experience more social rejection and devaluation than the general population.

The second aim of the current study was to test the hypothesis that higher shame would be associated with poorer socio-emotional functioning in young people with congenital dermatological disfigurement. This hypothesis was based on a number of studies confirming this relationship in non-disfigured adult and adolescent populations and some preliminary studies on shame and psychological functioning in young people with dermatological disfigurement.

1.2.3 Attachment, shame and psychological functioning.

Current models of psychological development propose that the developmental process involves many contributing variables that interact with each other in order to form unique developmental pathways for each individual. While there is theoretical and empirical support for main effects relationships between attachment and psychological outcome and shame and psychological outcome in people with congenital dermatological disfigurement, there is also evidence to suggest a relationship between attachment and shame, which will also be tested in this study. Despite a substantial theoretical literature that supports the relationship between attachment and shame, very little empirical research exists.

1.2.3.1 Attachment and Shame: Theoretical Evidence

As described in more detail in Section 1.2.2.2.3, many attachment and shame researchers view the two constructs as inextricably linked. (e.g., Bowlby, 1988; Lewis, 1971). Because the attachment relationship with the primary caregiver has been argued to be one of the most important socialising influences on the developing child (e.g., Belsky, Rovine, & Taylor, 1984), patterns of rejecting or devaluing behaviour from the primary attachment figure are said to result in excessive and unresolved shame responses which lead to the development of dispositional shame. Theorists argue for two potential pathways between attachment and dispositional shame: through the development of poor internalised emotional regulation via the lack of resolution to disruption to the attunement between attachment figure and infant (e.g., Schore, 1998) or via the development of negative internal working models of self as unvalued and others as rejecting (e.g., Bowlby, 1988). However, these hypothesised pathways, while theoretically compelling, are yet to be empirically demonstrated.

1.2.3.2 Attachment and Shame: Empirical Evidence

Despite such support from theorists there have been relatively few studies that have examined this relationship quantitatively. Only four empirical studies that investigate the link between attachment and shame have been published.

Lopez, Gover, Leskela, Sauer, Schirmer & Wyssmann (1997) conducted a study with 142 mainly female undergraduate university students. They assessed the relationships between attachment style using the Adult Attachment Style Questionnaire (AASQ; (Simpson, 1990; Simpson, Rholes, & Nelligan, 1992) and the Relationship Styles Questionnaire (RSQ; (Bartholomew & Horowitz, 1991) shame-proneness using the Test of Self-Conscious Affect (TOSCA; (Tangney, Wagner, &

Gramzow, 1989) and relationship problem-solving orientation using the Relationship Self-Efficacy Scale (RSES; (Lopez & Lent, 1991). The authors reported that attachment anxiety, as measured by the AASQ was significantly associated with shame-proneness (R^2= .24, p<.01). Using an ANCOVA, the study also found that the categorical measure of attachment style, as measured by the RSQ, significantly covaried with shame-proneness F(3,131) = 3.42, p<.02). Via a series of regression analyses, the authors found that while each shame-proneness (R^2= .38) and attachment style (R^2= .36) each significantly predicted relationship problem-solving style, when regressed together, with shame entered first and then attachment style, attachment style ceased to account for a significant proportion of the variance in the dependent variable leading the authors to conclude that the relationship between attachment and relationship problem-solving was mediated, in part, by shame.

Gross & Hansen (2000) sampled 204 university students and directly tested their hypothesis that attachment style (measured by the RSQ) and shame (Brief Shame Rating Scale; (Hibbard, 1994) were associated. Results supported their hypothesis indicating that secure attachment was significantly, inversely related to shame (R^2 = -.50, p<.001) and that fearful and pre-occupied attachment groups were each significantly associated with shame (R^2 = .27 and .26 respectively, p<.001). In contrast, dismissing attachment style was not significantly related to shame.

Gilbert et al (2009) in a study investigating the role of inferiority based striving evaluated striving, shame and attachment in 62 adults being treated for depression. The Experience of Close Relationships Questionnaire (Brennan et al., 1998) was used to assess attachment and the Other as Shamers scale (Goss et al., 1994b) was used to measure external shame. The study reported that shame and anxious attachment were significantly associated (r(62)=.68, p<0.01). The study also

reported that each external shame and anxious relationship style were independently and significantly associated with depression.

A study by Raque-Bogdan, Ericson, Jackson, Martin & Bryan (2011) investigated the relationship between attachment and self-compassion and "mattering", which are two constructs that can be considered to be the opposite to shame, in relation to mental and physical health outcomes. The study was conducted with a large sample of university students. As hypothesized, attachment security was significantly associated with self-compassion and mattering. While the study reported a non-significant relationship between attachment style and physical health, there was a significant association between attachment and mental health as well as between self-compassion and mattering and the physical and mental health outcome measures. In a meditational regression analysis, the authors found that self-compassion and mattering each significantly mediated the relationship between attachment style and mental health outcome.

1.2.3.3 Discontinuities between Attachment and Shame

While shame theorists are generally agreed that shame, as a normative emotion, has its origins in the primary attachment relationship, many theorists also concur that shame can be related to social experiences in broader interpersonal contexts and across the lifespan. These include controlling and/or critical parenting, experiences of explicit social devaluation, such as bullying, in the wider social context and stigmatization (See Section 1.2.2.2.3 for a more detailed account of causes of dispositional shame). As such, while it is hypothesised that shame would be associated with attachment security, shame could also vary according to other factors in the individual's life and should, therefore, reflect psychosocial influences beyond the attachment relationship.

In the case of children disfigured by their medical condition, the literature has suggested that there are several factors outside of the primary attachment relationship that may also increase the likelihood of developing dispositional shame: 1) general stigmatization (e.g., Masnari et al., 2013), the knowledge that being disfigured means that you are considered to be deviant or less valued than the general population, 2) experienced social rejection (e.g., Houston & Bull, 1994) such as direct bullying or more subtle forms of rejection such as other people being less likely to sit next to you in a public place and 3) altered parental and family relationships due to the child being disfigured and possibly also unwell in other ways related to the disfiguring medical condition. As such, while the effects of attachment and shame on psychological function may overlap, it is also expected that shame might also affect socio-emotional functioning independently of attachment.

1.3 The Current Study

The way in which attachment and shame, together, affect socio-emotional functioning in children and adolescents with congenital dermatological disfigurement has not yet been tested empirically. Based on current theories of the relationship between attachment and shame, as well as emerging empirical evidence on adult, non-disfigured populations, it is hypothesized that there will be a significant association between attachment and internal and external shame. It is also hypothesized that when attachment, internal and external shame are combined as independent variables, they will account for more variance in socio-emotional indices than each independent variable alone. That is, shame and attachment will account for some overlapping variance as well as each variable contributing independently to socio-emotional functioning.

1.3.1 The aims of the study.

The main purpose of this study was to understand why some children and adolescents with congenital dermatological disfigurement demonstrate positive social and emotional functioning while others experience significant difficulties in these domains.

To this end, the study aimed to develop an explanatory model of socio-emotional functioning in children and adolescents with congenital dermatological disfigurement. The explanatory model was based on the principles of prevailing developmental theory as a means of understanding how multiple factors might predispose a young person with congenital dermatological disfigurement towards idiosyncratic adaptive or maladaptive social and emotional functioning. It was hoped that identifying key factors influencing socio-emotional functioning in this population would contribute to developing better clinical services by improving the ability to identify individuals who might be more vulnerable to social and/or emotional difficulties and by helping to develop targeted psychological interventions that would increase therapeutic efficacy.

The current study aimed to utilise a cross-sectional design to conduct a preliminary exploration of the hypothesised relationships between congenital disfigurement, attachment, shame and socio-emotional functioning. While this design precluded the possibility of testing causality, it was hoped that positive findings might lead to a larger-scale study with a longitudinal design that would test the causal directions of the variables in the model.

Children and adolescents with congenital dermatological disfigurement were chosen as the target population for this study because they were considered to be under-represented in the existing empirical literature about disfigurement. Studies on

congenital disfigurement have focused on people with cleft lip and/or palate and, to a lesser degree, people with cranio-facial anomalies.

Social and emotional functioning were chosen as the psychological outcomes of the current study because the existing literature on young people with congenital disfigurement indicated that these were the domains of psychological functioning most likely to show deficits in this population. The current study aimed to test the hypothesis that children and adolescents with congenital dermatological disfigurement would experience more difficulties in socio-emotional functioning that the general population.

In the explanatory model, congenital dermatological disfigurement was conceptualized as a risk factor that resulted in social stress because of the negative way in which people perceive and respond to individuals with disfigured appearance, both in primary caring relationships and the wider social context. In the current study, two psychological factors emerged as potential candidates for contributing factors: attachment and shame.

Attachment was selected as a possible contributing factor because researchers have reported that congenitally disfigured children may be at risk of insecure attachment because their appearance may interfere with the way in which the child, in infancy, might interact with their primary attachment figure and vice versa (e.g., Wasserman & Allen, 1985) and because attachment style has been consistently shown to significantly contribute to socio-emotional functioning in non-disfigured populations (e.g., Sroufe, 2005). Preliminary evidence also supports this relationship in adults with dermatological conditions (e.g., Picardi et al., 2005)

In the current study, it was argued that attachment style would be particularly important to a disfigured child's socio-emotional development because this group of young people was more likely to experience negative social experiences due to their appearance. It was hypothesised that children with secure attachment would be able to tolerate these negative social experiences more successfully than insecurely attached individuals. This would mean that securely attached individuals would be protected from the negative effects of negative social experiences while insecurely attached young people would be more vulnerable to the negative effects of the negative social experiences which would ultimately lead to differential socio-emotional functioning within the population of young people with congenital dermatological disfigurement. It was also hypothesised that secure attachment might help to establish positive social relationships between the disfigured child and others, thereby potentially overriding the negative influence of their appearance on social interactions.

Shame was selected as a potential contributor to socio-emotional functioning in young people with congenital dermatological conditions. Because increased risk of psychological maladaptation in this population is based on the hypothesis that disfigured people experience more social rejection and devaluation than non-disfigured people, shame presented itself as a possible contributing variable.

The current study investigated two roles of shame in young people with congenital dermatological disfigurement: 1) young people with congenital dermatological disfigurement would report higher levels of internal and external shame because of the increased negative social experiences due to having a disfigurement; 2) dispositional shame would be associated with poorer socio-emotional functioning as has been found in studies with non-disfigured populations.

The current study also theorized that attachment insecurity would be associated with high shame because this relationship is supported by the theoretical and, more recently, empirical literature (e.g., Schore, 1998; Gross and Hansen, 2000). However, it was also hypothesized that shame would be affected by factors outside of attachment which would mean that shame would also have an independent effect on socio-emotional functioning. As such, attachment and shame would have an independent and overlapping relationship with socio-emotional functioning in this population. This explanatory model is illustrated in Figure 1.2. The aims and hypotheses of the current study are presented in Table 1.3.

Table 1.3
Study Aims and Hypotheses

	AIMS	**HYPOTHESES**
1	To describe the socio-emotional profile of young people with congenital dermatological disfigurement	Young people with congenital dermatological disfigurement will have higher levels of socio-emotional difficulty than the general population
2	To measure attachment style in young people with congenital dermatological disfigurement.	Young people with congenital disfigurement will be more likely to be insecurely attached than the general population.
3	To assess internal and external shame in young people with congenital dermatological disfigurement	There will be higher internal and external shame in young people with disfigurement than the general population.
4	To assess the association between attachment and shame in relation to socio-emotional functioning.	Attachment and shame independently and combined will be significantly associated with socio-emotional functioning.

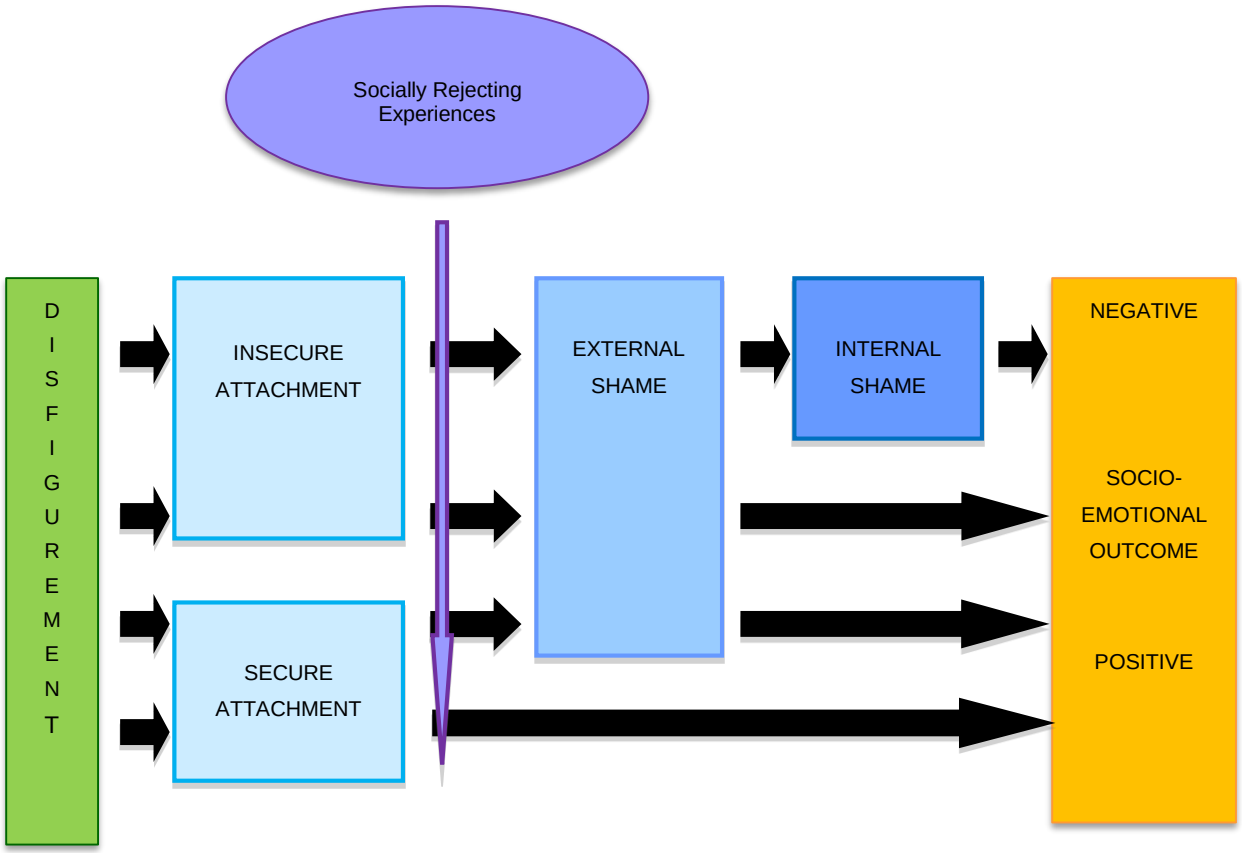

Figure 1.2. Proposed Explanatory Model of Socio-Emotional Functioning in Young People with Congenital Disfigurement

2.0

METHOD

2.1 Design

2.1.1 Study Design

The current study was designed as a cross-sectional investigation that included comparisons between the indexed sample and the general population as well as within subject analyses. A series of statistical analyses were utilized to test the hypotheses outlined in Table 1.3. The main analyses are described below.

Aim 1 set out to test the hypothesis that children with congenital dermatological disfigurement have higher levels of socio-emotional difficulty than non-disfigured children in the general population. This hypothesis was statistically tested by comparing mean scores from the outcome measures completed by the disfigured sample and their parent/guardian against published norms. Eight measures were used to assess socio-emotional functioning. These were subscales taken from the parent- and self-reported versions of the Strengths and Difficulties Questionnaire (SDQ; (Goodman, 1997) and the Harter Self-Perception Profile (SPP; (Harter, 1985; 1988) for Children and Adolescents. The measures were the SDQ parent- and self-reported Total Difficulties, Emotional Symptoms and Peer Problems subscales and the SPP Social Acceptance and Global Self-Worth subscales. Mean scores for each measure from the disfigurement group were compared against the published norms using a two-tailed single sample t-tests to test the hypothesis that the disfigured young people would have significantly higher social and emotional difficulties than the general population. The relationship between parent-reported and self-reported scores on the Total Difficulties, Emotional Symptoms and Peer Problems subscales of the SDQ were also analyses using a simple correlational analysis. Within-group differences relating to demographic and medical variables were also analysed using independent two-tailed t-tests for variables with two categories and analyses of variance (ANOVA) to test differences across more than two variable categories.

Aim 2 set out to test the hypothesis that congenitally disfigured children are significantly more likely than children in the general population to be insecurely attached. Attachment security was assessed using the Child Attachment Interview (CAI; (Shmueli-Goetz et al., 2008). Distribution across attachment classifications was compared between the sample group and published norms using a chi-square analysis.

Aim 3 set out to test the hypothesis that congenitally disfigured children would report significantly higher levels of external and internal shame than the general population. Because there are currently no published norms for children and adolescents for the shame measures used, data from a school-based comparison group were used to represent the general population (See Appendix E for details of comparison group data collection). External Shame was measured using the Others as Shamers Scale (OAS; (Goss et al., 1994b). Internal shame was measured using the Internalised Shame Scale (ISS; (Cook, 1988). A two-tailed t-test was used to compare disfigurement group and comparison group means on each measure.

Aim 4 set out to test the association between the independent variables: attachment, shame and the dependent variables: the measures of socio-emotional functioning. First, the relationship between each independent variable, separately, and socio-emotional functioning was investigated. A multivariate analysis of variance (MANOVA) was used to test the difference in socio-emotional functioning across secure and insecure attachment categorisations. A correlational analysis was used to assess the association between shame measures and socio-emotional functioning. Second, the relationship between attachment and shame was analysed by using a two-tailed t-test to compare the differences in shame scores between securely attached participants and insecurely attached participants. The combined

relationship between attachment, shame and socio-emotional functioning was then analysed using a series of hierarchical regression analysis.

At each of the four steps outlined above within group differences, based on medical and demographic indices, were investigated on all measures in order to identify any systematic patterns of variance in the data that might skew the main analyses.

2.1.2 Ethical Approval

Application for ethical approval for the current study was submitted to the Institute of Child Health/Great Ormond Street Hospital Research Ethics Committee in March, 2009 (REC reference number 09/H0713/19). A favourable ethical opinion was confirmed in April 2009 (Appendix F).

2.1.3 Power Calculation

Primary Power Calculation

The number of participants required to achieve adequate statistical power was calculated using GPower. The power calculation was based on the most complex statistical analysis in the current study: the hierarchical regression analysis with the independent variables of attachment and shame and the dependent variables consisting of measures of socio-emotional functioning.

There were few published studies that provided valid data on which to base the power calculation for the proposed multiple regression analysis. The primary power calculation was conducted using the results from (Irons & Gilbert, 2005) in which the association between attachment security, shame and depression, measured in an adult population, yielded a correlation of r =.5 and the association between shame and psychological outcome was r =.22. The association between attachment and shame remained the same at r =.25. The partial correlation between attachment

anxiety and depression controlling for shame was .47, and the R^2 partial was .222. The partial correlation between attachment and shame controlling for depression was .166, and the R^2 partial was .027. The partial correlation between shame and depression controlling for attachment was .113, and the R^2 partial was .013

On the basis of the second calculation, by adding shame into the hierarchical regression model, the explained variance increased by 13%. For this effect size, if alpha =.05, and power = 90%, the required sample size is 73 participants. This second sample size seemed more likely to capture key group differences than the original proposed sample size of 28. As such, the second calculated sample size was adopted for the current study.

In order to allow for multiple analyses, the Bonferroni corrected alpha was .05*2=.025, the effect size = .1494 and the power = 90% which increased the required sample size to 86.

Secondary Power Calculation

The main focus of the current study was to investigate within group associations, constituting three of the four key study aims. As such, the main sample size calculation was based on the relationships between the independent variables of attachment and shame and the dependent variable of socio-emotional functioning as described above. A further sample size calculation was conducted for the first study aim: to compare the disfigured sample with the general population on the indices of socio-emotional functioning. This second calculation was conducted to ensure that aim 1, also a substantive analysis, was also adequately powered.

In order to estimate the sample size required for adequate statistical power in the analysis of difference between the disfigurement group and the general population

on socio-emotional functioning, the current study looked to the findings of the similar studies included in the literature review reported in section 1.1.5. The reviewed studies presented a challenge in calculating sample size. First, none of these studies used a similar sample of young people with congenital dermatological disfigurement, all but two studies focusing on young people with CLP and cranio-facial anomalies. Second, the results of these studies were mixed with both positive, negative and no significant findings reported on the same and on different measures. Finally, there was a wide range of questionnaire measures used that measured a wide range of different psychological indices, some of which were relevant to the current study while others were not.

In order to determine an adequate sample size for Aim 1, two sample size calculations were conducted to represent the range of non-zero effect sizes reported i.e., one calculation to represent findings of significantly better socio-emotional functioning in the disfigured group and one calculation to represent findings of significantly worse socio-emotional functioning in the disfigured group. The calculations were also based on results directly relating to social and emotional functioning rather than the wider range of psychological indices included in the earlier relevant studies. Finally, the study with the most similar sample to the current study was identified.

The study by Sheerin et al (1995) was chosen to drive the power calculation for Aim 1 of the current study. The Sheerin study used a sample of young people with port-wine stains and prominent ears. The study also reported significantly better and significantly worse functioning in their disfigured groups compared to the general population. Finally, their significant results were in the relevant domain of social and emotional functioning. The index of social difficulties, measured by the Child behavior Checklist (Achenbach, 1991) found that the disfigurement group had

significantly worse social functioning than the general population. The index of global self-worth, measured by the Harter Self-Perception Profile (Harter, 1984), found that the port-wine stain group reported significantly better self-esteem than an age-matched school-based comparison group.

The effect size for social difficulties reported by Sheerin et al (1995) was calculated using the Cohen's *d* test for estimating sample size for t-test analyses resulting in the value of *d* =1.01445. Calculating sample size for independent samples, two-tailed t-test using *d* = 1.01445, α = 0.05 and statistical power = 0.8 yielded an estimated sample size of 17 participants per group.

The calculated effect size for global self-worth was *d* = 0.57445. Calculating the sample size for an independent samples, two-tailed t-test using *d* = 0.57445, α = 0.05 and power set at the default level of 0.8 yielded an estimated sample size of 49 participants per group.

As such, for the purposes of the current study, it was considered that a sample size of 49 or more would be sufficient for 80% power in the analysis of difference in socio-emotional functioning between the disfigured group and the general population.

As the estimated sample size to achieve adequate statistical power for the main regression analysis (N=86) was the biggest of the three sample sizes calculated, the study aimed to include at least 86 participants.

2.2 Sample

Participants in the disfigurement group were sampled from patients attending dermatology out-patient clinics at Great Ormond Street Hospital (GOSH), London,

who met the inclusion and exclusion criteria below. Shame data were also collected from a school-based comparison group. Information about the comparison group sample and procedure are in Appendix E.

2.2.1 Inclusion Criteria

- Participants were between the ages of 8 and 17 as this is the age range at which referrals for social and emotional difficulties peak. This is also the age range that is most under-represented in the relevant literature.
- Participants had congenital dermatological disfigurement, that is, disfigurement caused by a dermatological condition that has been present since birth to two years of age.

2.2.2 Exclusion Criteria

- Patients who were acutely, medically unwell were excluded in case their medical condition affected their concurrent mood state and, therefore, their responses to psychological measures. It was also considered to be unethical to disrupt the process of seeking medical intervention where the medical needs were acute.
- Patients who were attending GOSH for a major medical procedure as emotional distress or pre-medications related to their medical procedure might have affected their responses to the study measures.
- Patients with significant cognitive delay or any other significant developmental disorder, such as specific reading or sensory difficulties that might have impaired their ability to complete the data collection process.
- Patients who were not fluent and literate in the English language were excluded due to the requirement to read, write and speak English fluently. Furthermore, most of the measures used in this study were not yet normed for children who are not fluent in English.

2.2.3 Sample Selection

The full details of the sampling procedure are described in Section 2.4.1.

566 potential participants were identified from the dermatology out-patient clinic lists on the hospital patient information system and contacted by mail approximately two weeks prior to their next out-patient appointment. One week later, the potential participant was contacted by telephone by the researcher to provide more information about the study, if required, and to gain verbal consent for participation and to arrange a time and place to meet for data collection. One hundred and seventy two families (30.4%) could not be contacted by telephone, either because their telephone details were incorrect on the patient information system or because the researcher was unable to speak directly with the family prior to the scheduled out-patient appointment. Of the 394 families that were contacted, 91 did not participate because of scheduling difficulties (mainly because their dermatology appointment had been changed or because they did not have time, on the day of their appointment, to participate, 79 declined to participate because they were not interested in participating or because they were concerned about potential negative consequences of participating such as talking about their appearance being “too upsetting” for their child, 49 did not meet inclusion/exclusion criteria and in 46 cases, no reason for non-participation was recorded. A further seven participants were dropped from the study after meeting with the researcher because the researcher determined that they did not fit inclusion criteria. A total of 122 patients and their parents were included in this study. Nearly all participants were accompanied by their biological mother who completed the parent-reported measures. Some participants were accompanied by their mother and father. One participant was accompanied only by her biological father, who was her primary carer. One

participant was accompanied by her foster mother. The selection of participants from the sample population is represented in Figure 2.1.

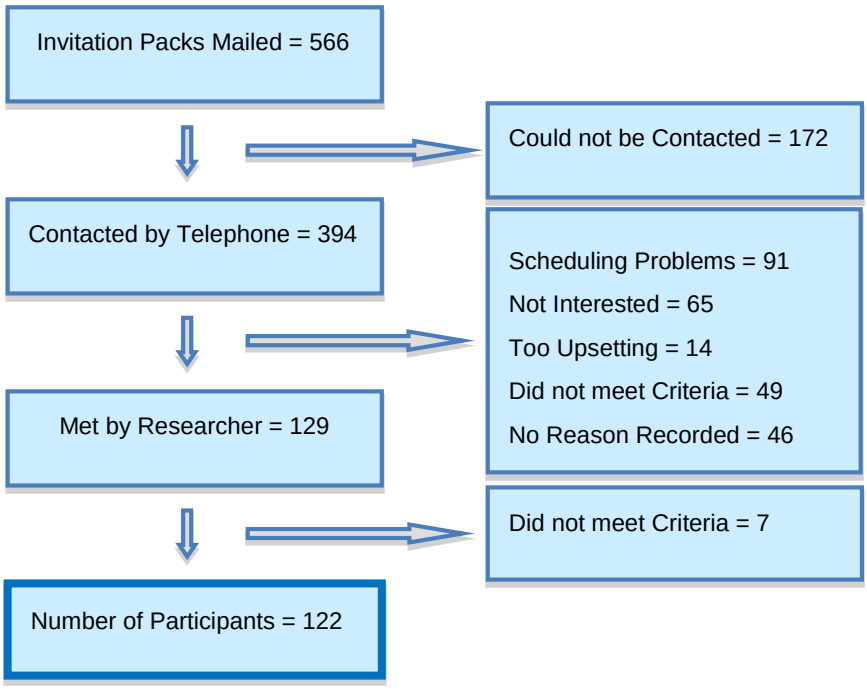

Figure 2.1. Selection of Participants from Sample Population

2.3 Measures

2.3.1 Descriptive measures.

2.3.1.1 Demographic Questionnaire (Appendix K)

A brief demographic questionnaire was constructed for this study. It identified the age of the child, the ethnicity of the child and the socio-economic status of the family using the self-rated version of the Standard Occupational Classification (National Office of Statistics, 2000). The questionnaire was completed by the accompanying parent/guardian.

2.3.1.2 Medical Condition Questionnaire (Appendix L)

The medical condition questionnaire was constructed for the purposes of this study and was completed by the parent/guardian. The measure asked for the patient's diagnosis, number of days spent in hospital in the last year, age of disease onset,

life threat, pain and mobility. To determine the location of disfigurement, parents/guardians were asked to mark its size and position on a simple diagram of a person (front and back). The medical conditions questionnaire.

2.3.2 Dependent variable measures.

The dependent variable was emotional and social functioning. The measures were eight subscales of two existing, well-constructed, widely used measures designed for children and adolescents; The Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997) and the Harter Self-Perception Profile (SPP)(Harter, 1985;1988). The SDQ subscales used were the parent-reported and self-reported Total Difficulties, Emotional Symptoms and Peer Relationship Problems subscales. The SPP subscales used were Social Acceptance and Global Self-Worth which were both completed by the child participant.

2.3.2.1 Strengths and Difficulties Questionnaire (SDQ): Total Difficulties, Emotional Symptoms and Peer Relationship Problems Subscales (Goodman, 1997)(Appendix Mi and Mii)

Three of the six subscales of the SDQ were used as measures of social and emotional functioning. These were Total Difficulties, Emotional Symptoms and Peer Relationship Problems. The parent-reported and self-reported versions of the SDQ were completed for all three subscales.

The SDQ is a popular and widely used measure of child and adolescent psychological functioning. The SDQ is considered to have good psychometric properties, making it a useful brief measure of the adjustment of children and adolescents. In a nationwide sample of 10, 438 British 5-15 year olds, SDQ data were obtained from parents, teachers, and children (Goodman, 2001). Analysis of this sample confirmed the predicted five-factor structure of the questionnaire and

found that it had satisfactory reliability (tested by internal consistency, cross-informant correlation and test-retest stability) and good validity.

The child self-report version of the SDQ was designed for completion by children from 11 years of age. However, a later study of its psychometric properties in a non-clinical 8-13 year old group (Muris, Meesters, Eijkelenboom, & Vincken, 2004) found that most psychometric properties of the questionnaire were satisfactory and comparable to those obtained from children in the over-11 age range.

Both parent report and child self-report versions of the included subscales were completed. As highlighted by De Los Reyes & Kazdin (2005) this multi-perspective approach is important in providing a more integrated understanding of adjustment in the population in question.

Each subscale consists of five items in the form of simple statements. For example, the Emotional Symptoms subscale elicits information about anxious and depressive features and includes items such as *"I am nervous in new situations. I easily lose confidence"*. The Peer Relationship Problems subscale includes items such as *"Other children pick on me or bully me"*. Responses are based on a three-point Likert scale ranging from "not true at all" to "very true".

The Total Difficulties subscale is the combined score of the Emotional Symptoms, Conduct Problems, Hyperactivity-Inattention and Peer Problems subscale scores. This subscale was used as a measure of overall psychological difficulties in the current study. The Total Difficulties subscale has been found to show the highest level of reliability and validity of all the subscales of the SDQ with internal reliability coefficients of 0.82 and 0.80 in the parent-report and child self-report versions respectively. Despite the relatively long period of time, of four-six months, over which to test test-retest reliability, the Total Difficulties subscale was found to have

acceptable test-retest reliabilities of 0.72 for the parent version and 0.62 for the child version. The inter-rater reliability of this subscale was significant at the $p<.001$ confidence level and higher than the meta-analytic mean. The Total Difficulties subscale also showed high construct validity when compared to DSM-IV psychiatric diagnoses (Goodman, Ford, Simmons, Gatward, & Meltzer, 2000).

The Emotional Symptoms subscale has been found to be a strong predictor of anxiety and depression in young people (Goodman, 2001) and was used, in the current study, to measure internalizing problems in the sample. The Emotional Symptoms subscale was found to have acceptable internal consistency with Cronbach Alpha coefficients of 0.67 and 0.66 for parent and child versions respectively and acceptable inter-rater correlation (r^2=0.35, $p<.001$) between parent and child report and acceptable retest stability of 0.57 for both versions after a four-six month interval.

The Peer Problems subscale was chosen as a measure of social functioning for the purposes of the current study. This subscale has also been found to have lower internal consistency with reliability coefficients of 0.57 and 0.41 for parent and child versions respectively. Inter-rater correlations were all significant with the parent-child correlation at r^2= .40 ($p<.001$). Retest stability was 0.61 and 0.54 for parent and child versions respectively (Goodman et al., 2000).

2.3.2.2 The Self-Perception Profiles: Social Acceptance and Global Self-Worth Subscales (Harter, 1985; 1988)(Appendix N)

Two sub-scales from the SPPC and its adolescent version, the SPPA, were used as measures of psychological functioning in the current study. These were Social Acceptance and Global Self-Worth.

The SPPC is a 36-item questionnaire developed for use with children aged 8-12. It is designed to evaluate children's perceptions of themselves in different functional domains. The scale consists of six independent subscales: Social Acceptance, Physical Appearance, Athletic Competence, Behavioural Conduct, Academic Ability and Global Self-Worth. Each subscale consists of six items. The questions are worded using a structured alternative format, which involves the child deciding which of two opposing statements are most like them, and then rating the degree of similarity by choosing "Really true for me" or "Sort of true for me". An example question is shown below:

Really true for me	Sort of true for me	Some kids find it *hard* to make friends	**BUT**	Other kids find it's pretty *easy* to make friends	Sort of true for me	Really true for me
☐	☐				☐	☐

Each question is scored from one to four, with a score of one representing low perceived competence and four representing high perceived competence. Half of the items are worded to begin with the positive description of self-worth, and the other half begins with a negative description of a lack of self-worth.

A number of studies of the psychometric properties of the SPPC have indicated that it is a reliable (as measured by internal consistency and test-retest stability) and a valid self-report measure for assessing children's self-esteem (e.g., Harter, 1985; Muris, Meesters, & Fijen, 2003).

The Self-Perception Profile for Adolescents (SPPA; Harter, 1988) was based on the SPPC, but developed so that the items were more relevant to an older population from 13 years of age upwards. This questionnaire has the same format as the

SPPC and measures perceived competence in the same domains with three additional domains that have particular relevance to adolescence – romantic appeal, close friendship and job competence. Each of the nine subscales contains five items. The content of the original six domains is paralleled across the child and adolescent questionnaires, with virtually identical wording for many items. Subscale scores are the mean of the items from each subscale. This allows the subscale scores across the child and adolescent versions to be compared.

The Social Acceptance subscale was used to evaluate social functioning in the current sample. The Social Acceptance subscale on both adolescent and child versions of the SPP is designed to elicit from the respondent, how liked or accepted he or she feels by their peers. This subscale includes items such as *"Some kids are always doing things with a lot of kids BUT other kids usually do things by themselves"*. The Social Acceptance subscale on both SPPC and SPPA had high internal consistencies of 0.78 and 0.82 respectively (Muris et al., 2003)

The Global Self-Worth subscale on the SPPC and SPPA is designed to evaluate the respondent's overall sense of their own worth or how much they like themselves in a way that was qualitatively separate from the other SPP subscales. The subscale includes items such as *"Some kids are very happy being the way they are BUT Other kids wish they were different"*. Mean internal consistency was 0.8 on the SPPC and 0.86 on the SPPA. (Muris et al., 2003).

2.3.3 Independent variable measures.

2.3.3.1 Child Attachment Interview (CAI; Shmueli-Goetz et al., 2008)

The CAI is an interview developed to assess how attachment patterns are manifested in middle childhood and adolescence (8-15 years). The development of the interview was based, conceptually and structurally, on both Ainsworth's Strange

Situation (Ainsworth et al., 1971) and the Adult Attachment Interview (AAI; (George et al., 1984). Infant attachment measures aim to trigger, then observe and measure, attachment behaviours. Adult attachment measures rely on the individual's own account of internalised attachment representations. Like the Strange Situation, the CAI was designed to place the participant under some relational stress by being interviewed by a stranger without a parent present. However, rather than observing attachment behaviours, the CAI then elicits the individual's internalised representations of the attachment relationship and caregiver availability more in keeping with the AAI. The CAI is designed to reveal variations in presentation of different attachment styles, but also be flexible enough to help the children to cope with the demands of the interview, whilst maintaining validity.

The reliability and validity of the CAI has been carefully assessed by the authors using a UK sample of 161 non-referred and 65 referred young people between seven and 12 years of age. Construct validity, internal consistency and inter-rater reliability were all reported to be high. Test-retest reliability was also reported to be high over three months but dropped to below 0.7 after 12 months which the authors attributed to normal shifts in attachment style over time in some individuals (Shmueli-Goetz et al., 2008; Target, Fonagy, & Shmueli-Goetz, 2003). The data from this set of studies also form the UK norms for the CAI used in the current study to represent the general population.

The interview consists of 18 questions (plus probe questions) that ask about the participant’s internal conceptualization of their relationship with their attachment figures. It elicits information about attachment behaviour, indirectly, by asking the participant to recall stressful relational events such as separations from their attachment figures, conflict between their attachment figures and experiences of threats to themselves such as through illness or injury. The CAI takes approximately

30-40 minutes to complete. The interview is video-recorded and transcripts are made of the interview. In discussion with the authors, the CAI was adapted for use with a congenitally disfigured population and made shorter to fit in with the practical demands of seeing children in a clinical context (see Appendix O). Seven items were removed from the original measure and replaced by one disfigurement-related item "What happens when someone is mean to you about the way that you look?" resulting in a measure consisting of 12 items.

The CAI is coded by watching the video recording and scoring the interview transcript simultaneously (See Appendix P for a sample transcript). The interviews are coded on eight scales: emotional openness, preoccupied anger, idealisation, dismissal, balance of positive/negative references to attachment figures, use of examples, resolution of conflicts, and overall coherence (See Appendix Q for coding form). While the CAI is still a relatively new measure and is still in development (personal communication from author), existing data indicate good psychometric properties, including inter-rater reliability, test-retest reliability, and concurrent and discriminant validities, in both clinical and non-clinical populations (Shmueli-Goetz et al., 2008). The measure yields a two-way classification of secure and insecure attachment to two or more attachment figures, usually the mother as the primary attachment figure and the father as the secondary attachment figure. However, participants are asked to independently identify their key attachment figures of which there may be more than one or two and who may not be their mother or father, for example, a step-parent, grand-parent or foster-parent. The CAI also yields three sub-categories of insecure attachment; dismissing, pre-occupied and disorganized attachment styles.

Three researchers completed formal CAI training and coding accreditation conducted by the authors of the measure at the Anna Freud Centre, the

international centre for CAI training. These researchers administered and coded the interviews. Inter-rater reliability, between the three coders, was calculated by assessing inter-rater agreement for the main classifications (secure, insecure-dismissing, insecure-preoccupied) on the first ten interviews, and was found to be high (classifications with respect to mother, kappa = 0.80; classifications with respect to father, kappa = 0.78). The researcher who administered the CAI to the participant did not code the interview to reduce bias. Each interview was coded by two researchers. Where there was disagreement between the two coders, or if there was any uncertainty about the coding, the third researcher also coded the interview.

2.3.3.2 Internalised Shame Scale (ISS; Cook, 1988)(Appendix S)

The ISS is a 30 item self report measure which measures negative global evaluations of the self with items reflecting feelings of inferiority, worthlessness, inadequacy, and alienation. Of the 30 items, there are 24 items with negative wording that capture shame, and six items with positive wording that capture self esteem. Clients respond on a 5 point Likert scale ranging from 0- "Never", to 4 – "Almost Always". A total ISS score is calculated by summing the item scores of the 24 shame related items, with total scores ranging from 0 to 96. Higher scores reflect higher internalised shame, with scores above 50 indicative of painful, possibly problematic, levels of internalised shame, and scores of 60 or above indicative of extreme levels of shame (Cook, 1996).

The ISS has demonstrated high internal consistency with both non- clinical (Cronbach's alpha = .95) and clinical (Cronbach's alpha = .96) adult samples (Cook, 1996). del Rosario and White (2006) examined test-re-test reliability and internal consistency on a large sample of university students. They found high internal consistency (Cronbach's alpha = 0.88) and high test-re-test reliability (r^2 = .81). The ISS has shown high internal consistency when used in research with adolescents

between 11 and 16 years of age (Cronbach's alpha = .94) (Soon & Harvey, 2009).

2.3.3.3 Other as Shamer Scale (OAS; (Goss et al., 1994a)(Appendix R)

This scale was devised as a modification of the Internalised Shame Scale (ISS; Cook, 1996) to explore expectations of how others see or judge the self rather than how the individual sees themselves. Thus, the focus is on "others see me as..". Items were chosen from the ISS and modified accordingly. For example, whereas the ISS asks the question "I feel like I'm never quite good enough", the OAS asks *"I feel other people see me as not good enough"*. Six items from the ISS were excluded because they did not lend themselves to this reversal of self-other evaluation, Thus the OAS described by Goss et al. (1994) was an 18-item scale. Following recommendations from the Institute of Child Health/Great Ormond Street Hospital Research Ethics Committee, the wording of some items was altered to make it more accessible to a younger audience and eight positively worded dummy items were added to ameliorate the impact of the largely negative focus of the questionnaire items. These additional eight dummy items were excluded from the scoring. One original item was removed from the original OAS because factor analysis has shown that it did not load onto any of the three identified factors (Goss et al., 1994). Therefore, the scale consisted of 25 descriptions of feelings and experiences. Participants respond on a 5-point Likert scale ranging from 1- "Never" to 5 – "Always". A total OAS score is calculated by summing item scores, with higher scores reflecting higher levels of externalised shame. The maximum score is 85 and the minimum 25.

The OAS has shown high internal consistency with adults (Cronbach's alpha = .92; Goss et al., 1994) and adolescents aged 11 to 16 years of age (Cronbach's alpha = .93)(Soon & Harvey, 2009).

2.4 Procedure

The data were collected by a research group of five doctoral researchers and three part-time honorary assistant psychologists, led by the current author over a seven-month period. The way in which the combined data collection was organised is described below.

2.4.1 Research Group

The data for the current study were collected by a team of researchers, some of whom used subsets of the same data in separate doctoral dissertations. The current author (KS) was the named Chief Investigator on all Research and Development Department and National Research Ethics Committee registration documentation and acted as supervisor and coordinator of the other members of the research group. The group consisted of four Doctorate in Clinical Psychology (DClinPsy) trainees (AS, JG, RM, KH) and three part-time honorary psychology assistants (SM, EH, MP).

The DClinPsy trainees used subsets of the data reported in the current study for their own dissertations although each study used common and unique data sets to test different research questions (See Appendix G for a more detailed description of the other doctoral studies completed alongside the current study). Data that have not been reported in the current study were collected alongside the data for the current study for the purposes of the other DClinPsy studies (See Appendix H for a table of measures used for each dissertation). Standards of conceptual originality and independence of contribution were applied according to the "Guidance to Undertaking a PhD while Supervising the Research of Professional Doctorate Students " of the Professorial Committee of the Research Department of Clinical, Educational and Health Psychology, University College London (Appendix I).

Declarations, written by each UCL researcher and included in each dissertation that these guidelines were followed, are shown in Appendix J.

Honorary assistant psychologists were involved in supportive duties including identifying clinic patients to be contacted, contacting schools in regard to collecting the comparison group data, maintaining database records and entering data.

2.4.2 Sampling.

- Participants were identified from the hospital patient information management system (PIMS)
- PIMS was checked each week for patients scheduled to attend dermatology out-patient appointments in two weeks time.
- Patients were checked for the inclusion/exclusion criteria of age and acute medical procedures on the day of their appointment.
- Patients fulfilling inclusion criteria, so far, and their parents/guardians were mailed information sheets about the study; one written for the child patient (Appendices T, U) and one information sheet written for the parent or guardian (Appendix V). The information sheets were accompanied by a standard letter inviting the patient and parent or guardian to participate in the study (Appendix W).
- The mailed information pack was followed up with a telephone call by a researcher to provide more information about the study, if required, to gain verbal consent for participation and to arrange a time and place to meet for data collection which coincided with their upcoming dermatology out-patient appointment.
- An Excel® spreadsheet was created to record contacts with potential participants in order that the researchers were able to work collaboratively and

to ensure adherence to the approved procedure. The contacts spreadsheet was saved onto a secure, networked drive separately to the spreadsheet of anonymised data.

2.4.3 Data collection.

- Data were collected at Great Ormond Street Hospital either in a consulting room near to the patient's scheduled out-patient appointment or in the Somers Clinical Research Facility at Great Ormond Street Hospital.
- The researcher met with the potential participant and their parent or guardian as agreed in the initial telephone conversation.
- At this point, information about the study was re-iterated and written consent was sought from the parent or guardian and signed assent was sought from the child patient (See Appendix X and Y for consent and assent forms respectively).
- Once the consenting process was completed, each child participant was asked to complete a standard test battery which took between 20 and 45 minutes to complete.
- The adult accompanying the child patient completed the parent-reported measures which usually took up to 15 minutes to complete.
- A researcher was present at all times to support the completion of the questionnaires. The parent and child were instructed to complete their questionnaires independently of one another.
- Once the questionnaires were completed, the parent was asked to leave the consultation room and to wait in the nearby waiting area.
- The CAI was then completed by the researcher and the child on their own. The CAI took between 20 minutes and 45 minutes to complete.
- Once the CAI was completed, the child participant was returned to their accompanying parent and both participants were given the opportunity to

discuss the experience of participating in the study and to ask questions about the process to the researcher.

2.4.4 Data entry

- Once the data had been collected from a participant, the anonymised questionnaire data were entered onto an SPSS datafile.
- The CAI video recordings were transcribed by one of the researchers.
- The CAI video recordings were saved onto a single external hard drive that was stored according to the Data Protection Act (1998). The interview transcripts were saved in an electronic datafile and in hardcopy also according to the data protection requirements.
- Each CAI recording was then coded by two accredited CAI coders. A third accredited coder reviewed interviews when the two primary coders were unable to agree an attachment classification or where there was any uncertainty about the coding.
- Coding information was saved onto the SPSS datafile and in hardcopy.

3.0

RESULTS

3.1 Sample Characteristics

Demographic and medical information about each participant was provided by the parent or guardian who accompanied the child participant. Socio-economic status was assessed using the occupational rating from the National Office of Statistics, Socio-Economic Classification, Self-Coded Measure and the occupation of the primary income earner in the participant's household (Nos, 2012). Details of the sample characteristics are provided in the tables below.

3.1.1 Demographic information.

The disfigurement group had a mean age of 12 years and two months, ranging from 8 years and 0 months to 16 years and 11 months of age. Sixty eight participants were secondary school students constituting a slight majority at 57.6% of the sample. Fifty participants were in primary school (42.3%). Nearly two thirds of the sample was female. Eighty-six percent of the sample identified themselves as white. Only 9.7% of the sample identified themselves as non-white and data for ethnicity on the remaining 4.2% was missing. The representation of white versus non-white participants in the current sample is consistent with the UK national population of which 86% are reported to be white and 14% non-white (NOS, 2012) Over 50% of the sample described the primary income earner as working in professional or managerial roles. The demographic information is summarised in Table 3.1.

Table 3.1
Demographics

Age/School	
Mean Age	146 mths (12yrs, 2m)
Age Range	8yrs 0m - 16yrs 11m
Primary School	50(42.3%)
Secondary School	68(57.6%)
Gender	**n (%)**
Female	74 (60.7)
Male	48 (39.3)
Ethnicity Category	**%**
White British	73.8
White Other	12.3
Mixed Race	4.9
Asian	3.2
Black	1.6
Other (inc. Arab)	0
Unknown	4.2
Occupational Category	**%**
Higher Professional/Management	29.6
Lower Professional/Management	24.6
Technical/Craft	9.8
Lower Supervisory	9.0
Semi routine	8.2
Intermediate	3.3
Routine	5.0
Unemployed (including benefits)	5.7
Unclassified	4.8

3.1.2 Medical Information

Nearly half of the sample (43.4%) had a vascular birthmark. The next most common dermatological condition was eczema at 18% of the sample. Over half of the sample (59.8%) reported that the dermatological condition manifested itself on their face. Over 28% of the sample was affected on other parts of their body only, and not on their face. Nearly 70% of the sample attended hospital appointments up to twice a year indicating that their dermatological conditions were fairly well controlled while 11.4% of the sample attended appointments six or more times per year suggesting a more complex and troublesome dermatological condition. Information about the disfiguring dermatological conditions affecting the current sample are summarised in Table 3.2.

Approximately 30% of the sample reported experiencing pain, mobility problems or other co-morbid medical problems such as asthma. There was a high level of

overlap between disease factors. Many participants who reported co-morbid pain also reported mobility problems and other illnesses. For 17.2% of the sample, their dermatological condition was potentially life threatening. The information about other medical features of the disfiguring dermatological condition are summarised in Table 3.3. These disease variables suggested a broad range of complexity of dermatological condition in the sample. Descriptions of the most common dermatological conditions represented in this sample are provided in Appendix Z.

Table 3.2
Dermatological Condition

Grouped Condition	n (%)
Vascular Birthmarks	53 (43.4)
Eczema	22 (18)
Bullous Disorder	13 (10.7)
Overgrowth Syndrome	8 (6.6)
Naevii	11 (9.0)
Other	8 (6.6)
Mastocytoses	7 (5.7)
Location of Disfigurement	**n (%)**
Face only	36 (29.5)
Face and other parts of body	37 (30.3)
Limbs only	28 (23.0)
Torso only	7 (5.7)
Unknown (missing data)	14 (11.5)
Age of Onset	**n(%)**
Birth	98 (80.3)
Birth - 24 mths	24 (19.7)
Number of Hospital Visits	**n (%)**
0-2	77 (67.6)
3-5	24 (21.1)
6 or more	13 (11.4)

Table 3.3
Other Medical Features

	Yes n (%)	No n (%)	Unknown n (%)
Pain	32 (26.2)	86 (70.5)	4 (3.3)
Mobility problems	36 (29.5)	79 (64.7)	7 (5.7)
Other medical problems	34 (27.9)	83 (68.0)	5 (4.1)
Potentially fatal	21 (17.2)	89 (73.0)	12 (9.8)

3.2 Aim 1: Socio-Emotional Functioning

The first aim of the current study was to compare the group of young people with congenital dermatological disfigurement against the general population on measures of socio-emotional functioning. The eight measures of socio-emotional

functioning used were the Total Difficulties, Emotional Symptoms and Peer Relationship Problems subscales from the parent-reported and self-reported SDQ and the Social Acceptance and Global Self-Worth subscales from the Harter SPP.

Aim 1 tested the hypothesis that children and young people with congenital dermatological disfigurement would be significantly worse off than the general population on all indices of socio-emotional functioning.

As a subsidiary aim, within group differences in socio-emotional functioning, based on key demographic and medical variables, were also investigated.

3.2.1 Analytic strategy.

The hypothesis was tested by comparing the disfigurement group mean for each socio-emotional outcome measure against British general population means for 4-15 year olds published for the SDQ subscales (Sdqinfo, n.d.) and norms published by Harter (1985; 1988) for the SPP subscales using a single-sample t-test.

All outcomes measures were tested for normality of distribution before means comparison analysis. Skewness and Kurtosis were checked for each measure. The five self-reported measures were all normally distributed. The three parent-reported measures showed a negative skew. Because the majority of measures, overall, showed a normal distribution and because the standard means of transforming data might not successfully address the skewness in some of the data, it was decided not to transform the data (Field, 2005).

A two-tailed chi-squared test of variance was also conducted on each group mean in order to determine if variance in scores differed across groups. A two-tailed test was chosen as there were no hypotheses about direction of inequality.

A series of group means comparison analyses were conducted to test differences in scores across demographic and medical variables. The demographic variables were gender and school stage. The medical variables were disfigurement location, dermatological diagnosis, mobility problems, co-morbid medical problems, life threat and pain. Binary variables were tested using an independent samples two-tailed t-test. Variables with more than two conditions were tested using an analysis of variance (ANOVA).

3.2.2 Findings

Table 3.4 summarises the data generated by the t-test comparison between the disfigurement group mean scores and the published norms on socio-emotional functioning measures.

Table 3.4
Socio-Emotional Functioning: Mean Scores

	Parent Report		Self-Report	
	Disfigured	Norm	Disfigured	Norm
SDQ Subscales	M (SD)	M (SD)	M (SD)	M (SD)
Total Difficulties	8.7 (5.9)	8.4 (5.8)	10.4 (5.9)	10.3 (5.2)
Emotional Symptoms	2.8 (2.5)	1.9 (2.0) ***	2.9 (2.2)	2.8 (2.1)
Peer Problems	1.6 (1.9)	1.5 (1.7)	1.8 (1.9)	1.5 (1.4)
SPP Subscales			M (SD)	M (SD)
Social Acceptance	N/A	N/A	3.24 (.56)	2.90 (.68) ***
Global Self-Worth	N/A	N/A	3.16 (.63)	3.03 (.64)*

****p<.001, **p<0.1, *p<0.5*

Parents reported significantly higher levels of Emotional Symptoms in the disfigured group compared to the normal population t(df=1,114)=3.801, p<.001. The child participants reported significantly better social acceptance t(df=96)=5.97, p<.000 and global self-worth t(df=101)=2.12, p=.037 than the published mean scores for the general population. There were no other significant differences between the disfigured group and the general population.

The chi-squared test of group variances found that variance in the disfigurement group and the general population was not significantly different on all measures

except for the measure of self-reported social acceptance. The social acceptance scores indicated that there was significantly greater variance in the general population on this measure yielding a score of T=65.12 which was lower than the critical value of $\chi^2(96)=70.78$.

In addition to comparing the mean scale scores of the disfigured group with the general population norms, the distribution of responses across the established clinical categories of the SDQ subscales was also compared. The SDQ is constructed in such a way that 80% of the general population fall into the normal range, 10% in the "borderline" range, and 10% in the "abnormal" range.

The percentages of disfigured participants falling into borderline and abnormal ranges according to both self and caregiver reports are shown in Table 3.5. Binomial probability tests were conducted to determine if the disfigurement group showed a significantly different likelihood of placement in any of the clinical categories.

Table 3.5
Percentage of the sample in the clinical ranges on SDQ subscales

	Parent-report		Self-report	
	borderline	abnormal	borderline	abnormal
Total Difficulties	12.9	9.5	16.2*	7.2
Emotional Symptoms	8.6	23.3**	5.4	7.2
Peer Problems	7.8	18.2	9	6.3

Based on parental report, these percentages suggested that the children in the sample were significantly more likely than the general population to score in the abnormal range on Emotional Symptoms (using a binomial probability test, $p=.002$). The disfigured group were significantly more likely to score in the borderline clinical range on self-reported Total Difficulties than the general population (using a binomial probability test, $p=.02$ for total score). There was no increased risk of caseness on the other SDQ subscales.

There were three outcome variables that were rated by both parent and child; SDQ Total Difficulties, Emotional Symptoms and Peer Problems. A two-tailed Pearson's Correlational Analysis was used to assess the association between parent and child ratings on each variable. Parent and child ratings on Total Difficulties ($r(109)=.573$), Emotional Symptoms ($r(109)=.534$) and Peer Problems ($r(109)=.581$) were found to be significantly correlated at an alpha level of .01. Parent-child report correlations are presented below alongside published correlations (Goodman, 2001) demonstrating that the parent and child reported scores in the current study were higher than the published correlations.

Table 3.6
Parent and Child Reported SDQ Subscales

	Total Difficulties	Emotional Symptoms	Peer Problems
Current Study	.57	.53	.58
Published *r*'s	.48	.37	.44

The results indicated a moderate correlation between parent and child reported scores on the Total Difficulties, Emotional Symptoms and Peer Problems subscales of the SDQ. Prima facie comparison with general population scores indicated that parent and child reported scores were more similar in the current study than in the general population. However, the differences in correlations in the general population and in the disfigured group were not statistically analysed for significance.

In summary, there was limited support for the hypothesis that the disfigurement group would experience significantly poorer socio-emotional functioning than the general population with mixed findings recorded. The disfigured group scored significantly higher on only one parent-report measure: the Emotional Symptoms subscale of the SDQ and the disfigured group were more likely to be scored in the

abnormal range on this subscale than the general population. The disfigured group was significantly more likely to be in the borderline clinical range on the self-reported Total Difficulties subscale despite no significant differences in mean scores. However, the disfigured group also self-reported significantly better social acceptance and global self-worth than the general population. It should be noted that there were a number of significant within-group differences based on disfigurement and medical variables. These within-group differences suggested that the disfigurement group might be quite heterogeneous in terms of illness variables and their influence on socio-emotional functioning.

A further set of analyses were conducted to make within-group comparisons in the group of young people with congenital dermatological disfigurement on the demographic variables of age and gender and the medical variables of medical diagnosis, disfigurement location, and the presence of the co-morbid symptoms of pain, physical disability and the presence of any co-morbid medical conditions such as asthma.

A comparison of socio-emotional functioning by gender yielded only one within group difference with parents rating boys as having higher Total Difficulties (M=10.76) than girls (M=7.45), $t(114) = -3.05$, $p = .003$.

Table 3.7
Gender

	Gender				
	Boys N=42	Girls N=69	*t*	df	*p*
Child-rated	M(SD)	M(SD)			
Total Difficulties	11.21(5.68)	9.97(6.04)	-1.08	109	.285
Emotional Symptoms	3.10(2.17)	2.71(2.29)	-.88	109	.383
Peer Problems	2.12(2.17)	1.59(1.66)	-1.43	109	.153
Social Acceptance	3.21(.69)	3.25(.48)	.30	95	.762
Global Self-Worth	3.22(.60)	3.13(.65)	-.683	100	.496
Parent-Rated					
Total Difficulties	10.76(5.54)	7.45(5.79)	-3.05	114	.003**
Emotional Symptoms	3.29(2.46)	2.46(2.50)	-1.74	114	.084
Peer Problems	2.04(2.04)	1.59(1.66)	-1.90	114	.060

Age was categorized into two groups: primary school age and secondary school age. Peer problems were reported by the child participants to be significantly worse in primary school (M=2.17) compared to secondary school participants (M=1.42), *t*(108) = 2.22, *p*=.029 (Table 3.8). Self-reported Emotional Symptoms approached significance with the primary school-aged participants reporting a slightly higher level of difficulty in this domain. All other socio-emotional variables were not significantly different according to school stage.

Table 3.8
School Stage

	School Stage		*t*	df	*p*
	Primary N=46	Secondary N=64			
Child-rated	M (SD)	M (SD)			
Total Difficulties	11.24(5.71)	9.63(5.72)	1.46	108	.147
Emotional Symptoms	3.28(2.16)	2.48(2.22)	1.88	108	.06
Peer Problems	2.17(2.00)	1.42(1.55)	2.22	108	.029*
Social Acceptance	3.21(.60)	3.28(.53)	-.60	94	.549
Global Self-Worth	3.17(.66)	3.16(.61)	.08	99	.937
Parent-Rated					
Total Difficulties	9.33(.68)	8.27(5.19)	.94	113	.347
Emotional Symptoms	3.00(2.65)	2.59(2.40)	.86	113	.389
Peer Problems	1.47(1.85)	1.71(1.83)	-.70	113	.485

The variable of disfigurement location significantly differentiated the participants on self-reported peer problems *F*(7,103)= 2.753, *p*=.012 and parent-reported peer problems *F*(7,108)=3.427, *p*=.002 and total difficulties *F*(7,108)=3.557, *p*=.002. An LSD post-hoc analysis revealed that the participants who were disfigured on their whole body, face and torso and limbs and torso reported significantly worse scores on these three subscales than the participants who were affected on their face only, limbs only and face and limbs. The results indicated that the participants who had greater areas of their body affected by the disfigurement scored worse than those participants who had smaller areas of body affected. This appeared to be the case regardless of the location of the disfigurement. However, due to the notable differences in group sizes, with only four participants in the Face and Limbs group

compared to 31 participants in the Face Only group, these findings should be considered with caution. A further analysis of Disfigurement Location data constituted the Post Hoc Study in which the visibility of the disfigurement is used as the basis for comparing psychosocial data.

Table 3.9
Disfigurement Location: Mean Scores

	Whole Body N= 24	Face & Limbs N= 4	Limbs & Torso N= 8
Child-rated	M (SD)	M (SD)	M (SD)
Total Difficulties	12.33 (5.84)	6.75 (2.99)	11.50 (6.3)
Emotional symptoms	3.38 (2.08)	1.0 (.0)	2.88 (2.59)
Peer problems	2.54 (2.57)	1.25 (.5)	2.63 (1.5)*
Social acceptance	3.0 (.79)	3.6 (.37)	3.33 (.34)
Global Self-Worth	3.11 (.56)	3.53 (.26)	3.04 (.59)
Parent-Rated			
Total Difficulties	12.08 (5.62)**	8.75 (2.22)	11.00 (9.97)
Emotional Symptoms	3.62 (2.26)	2.00 (1.41)	4.25 (3.41)
Peer Problems	2.65 (2.23)**	1.75 (2.21)	1.88 (2.03)

Table 3.9 continued
Disfigurement Location: Mean Scores

	Face Only N= 31	Limbs Only N= 26	Torso Only N= 9
Child-rated	M (SD)	M (SD)	M (SD)
Total Difficulties	8.94 (5.35)	9.92 (5.66)	9.67 (7.43)
Emotional symptoms	2.39 (2.39)	2.62 (1.81)	3.33 (1.8)
Peer problems	1.26 (1.26)	1.54 (1.66)	1.56 (1.42)
Social acceptance	3.29 (.45)	3.19 (.58)	3.53 (.44)
Global Self-Worth	3.29 (.58)	3.14 (.66)	3.25 (.67)
Parent-Rated			
Total Difficulties	6.42 (5.06)	7.78 (4.95)	5.11 (3.86)
Emotional Symptoms	2.06 (2.38)	2.52 (2.42)	1.67 (2.06)
Peer Problems	.88 (1.27)	1.30 (1.54)	.89 (1.36)

Table 3.10
Disfigurement Location: F Scores

	F	df	*p*
Child-rated			
Total Difficulties	1.77	7, 103	.101
Emotional symptoms	1.952	7, 103	.069
Peer problems	2.753	7, 103	.012*
Social acceptance	1.191	7, 89	.316
Global Self-Worth	1.251	7, 94	.283
Parent-Rated			
Total Difficulties	3.557	7, 108	.002**
Emotional Symptoms	2.083	7, 108	.051
Peer Problems	3.427	7, 108	.002**

The variable of dermatological diagnosis significantly differentiated the socio-emotional factors of self-rated peer problems $F(7,103)= 2.753$, $p=.012$ and parent-rated Total Difficulties $F(7,108)= 3.557$, $p=.002$ and Peer Problems $F(7,108)= 3.427$, $p=.002$ while parent-rated Emotional Symptoms was approaching significance $F(7,108)= 2.083$, $p=.051$. A Bonferroni post-hoc analysis found that the atopic eczema group and the overgrowth syndromes group scored significantly worse on parent-rated Total Difficulties than the Vascular Tumours group and that the Overgrowth Syndrome group scored significantly worse on Self-rated Peer Problems and Parent-rated Total Difficulties, Emotional Symptoms and Peer Problems than the Vascular Tumours group.

Table 3.11
Dermatological Diagnosis: Mean Scores

	Atopic Dermatitis N= 12	Vascular Tumours N= 49	Bullous Disorders N=13
Child-rated	M (SD)	M (SD)	M(SD)
Total Difficulties	11.74 (6.91)	9.00 (5.25)	11.83(5.41)
Emotional Symptoms	3.32 (2.36)	2.57 (2.11)	3.08(2.15)
Peer Problems	2.16 (2.24)	1.16 (1.09)	2.25(2.3)
Social Acceptance	3.20 (.72)	3.30 (.40)	3.04(.81)
Global Self-Worth	3.10 (.59)	3.18 (.61)	3.07(.85)
Parent-Rated			
Total Difficulties	12.90 (5.9)***	6.64 (5.06)	9.15(3.98)
Emotional Symptoms	3.50 (1.96)	2.14 (2.21)	2.92(2.22)
Peer Problems	2.60 (2.09)	.88 (1.19)	1.77(1.92)

Table 3.11
Dermatological Diagnosis: Mean Scores continued

	Overgrowth Syndromes N= 6	Mastocytoses N=7
Child-rated	M (SD)	M (SD)
Total Difficulties	13.33 (8.71)	9.00 (6.00)
Emotional Symptoms	3.67 (2.66)	2.14 (2.41)
Peer Problems	3.33 (3.20)*	1.71(1.11)
Social Acceptance	2.59 (.30)	3.32 (.72)
Global Self-Worth	3.07 (.77)	3.53 (.50)
Parent-Rated		
Total Difficulties	12.43 (6.997)	10.71(8.1)
Emotional Symptoms	5.43 (3.26)**	3.43 (3.74)
Peer Problems	3.14 (2.41)***	1.86 (2.12)

Table 3.12
Dermatological Diagnosis: F Scores

	F	df	*p*
Child-rated			
Total Difficulties	1.308	6, 104	.260
Emotional Symptoms	.599	6, 104	.730
Peer Problems	2.601	6, 104	.022*
Social Acceptance	1.596	6, 90	.157
Global Self-Worth	.527	6, 95	.786
Parent-Rated			
Total Difficulties	5.145	6, 109	.000***
Emotional Symptoms	3.438	6, 109	.004**
Peer Problems	6.205	6, 109	.000***

Participants with mobility problems had significantly poorer functioning on the measures of self-reported Total Difficulties, Self-reported Peer Problems and all three parent-reported measures of Total Difficulties, Emotional Symptoms and Peer Problems.

Table 3.13
Mobility

	Problems N= 35	Normal N= 75	*t*	df	*p*
Child-rated	M (SD)	M (SD)			
Total Difficulties	12.23 (6.39)	9.64 (5.56)	2.167	108	.032
Emotional Symptoms	3.37 (2.51)	2.64 (2.09)	1.602	108	.112
Peer Problems	2.51 (2.16)	1.48 (1.64)	2.514	52.949	.015*
Social Acceptance	3.07 (.68)	3.32 (.48)	-1.850	46.807	.071
Global Self-Worth	3.17 (.65)	3.16 (.62)	.058	99	.954
Parent-Rated					
Total Difficulties	11.34 (6.27)	7.53 (5.32)	3.313	112	.001**
Emotional Symptoms	3.71 (2.57)	2.35 (2.39)	2.736	112	.007**
Peer Problems	2.49 (2.17)	1.20 (1.52)	3.165	49.356	.003**

The presence of chronic or recurrent pain was associated with significantly poorer socio-emotional functioning on all measures except for self-reported global self-worth. The results are shown in Table 3.14.

Table 3.14
Pain

	Pain N= 29	No Pain N= 81	*t*	df	*p*
Child-rated	M (SD)	M (SD)			
Total Difficulties	12.72 (6.23)	9.65 (5.64)	2.445	108	.016*
Emotional Symptoms	3.62 (2.38)	2.60 (2.15)	2.123	108	.036*
Peer Problems	2.69 (2.24)	1.49 (1.63)	2.638	39.137	.012*
Social Acceptance	2.95 (.73)	3.34 (.46)	-2.516	31.06	.017*
Global Self-Worth	3.07 (.68)	3.19 (.61)	-.890	99	.376
Parent-Rated					
Total Difficulties	12.67 (6.34)	7.27 (5.05)	4.695	113	.000***
Emotional Symptoms	3.77 (2.65)	2.41 (2.37)	2.609	113	.010**
Peer Problems	2.73 (2.56)	1.21 (1.48)	3.439	38.168	.001***

Participants who were reported to have co-morbid medical problems had significantly poorer scores on self-rated Total Difficulties and Parent-rated Total Difficulties, Emotional Symptoms and Peer Problems. The difference in Self-rated Peer Problems was approaching significance with the co-morbid group reporting more difficulties in this area.

Table 3.15
Medical Co-morbidity

	Yes N= 31	No N= 79	*t*	df	*p*
Child-rated	M (SD)	M (SD)			
Total Difficulties	12.58 (6.69)	9.63 (5.43)	2.39	108	.018*
Emotional Symptoms	3.42 (2.47)	2.66 (2.13)	1.61	108	.110
Peer Problems	2.48 (2.41)	1.54 (1.56)	2.01	40.275	.051
Social Acceptance	3.08 (.63)	3.30 (.53)	-1.69	94	.095
Global Self-Worth	3.11 (.53)	3.18 (.66)	-.45	99	.657
Parent-Rated					
Total Difficulties	11.58 (5.17)	7.51 (5.79)	3.51	113	.001***
Emotional Symptoms	3.70 (2.00)	2.39 (2.60)	2.59	113	.011*
Peer Problems	2.48 (2.25)	1.26 (1.51)	2.88	44.124	.006***

Significant differences were found on the measures of self-rated Peer Problems and parent-rated Total Difficulties and Emotional Symptoms. A Bonferroni Post Hoc analysis revealed that participants with a potentially life threatening dermatological condition reported significantly worse peer problems than the participants whose medical condition was not life threatening. The group who were unsure about whether their medical condition was life-threatening or not scored significantly worse than both other groups on parent-rated Total Difficulties and Parent-rated Emotional

Symptoms. However, because this group only had three members, these findings are not easily interpreted or generalized.

Table 3.16
Life Threat: Mean Scores

	No Life Threat N=85	Life Threat N=19	Don't Know N=3
Child-rated	M (SD)	M (SD)	M (SD)
Total Difficulties	10.27 (5.61)	11.11 (7.61)	11.67 (8.33)
Emotional Symptoms	2.76 (2.13)	3.42 (2.78)	3.33 (3.22)
Peer Problems	1.60 (1.61)	2.89 (2.71)*	2.00 (1.00)
Social Acceptance	3.24 (.55)	3.19 (.65)	3.25 (.12)
Global Self-Worth	3.15 (.66)	3.13 (.53)	3.42 (.12)
Parent-Rated			
Total Difficulties	7.98 (5.59)	10.45 (5.67)	16.00 (10.44)*
Emotional Symptoms	2.31 (2.22)	4.00 (2.51)	6.00 (4.00)**
Peer Problems	1.34 (1.66)	2.70 (2.27)	2.67 (1.53)

Table 3.17
Life Threat: F Scores

	F	df	*p*
Child-rated			
Total Difficulties	.209	2, 104	.812
Emotional Symptoms	.703	2, 104	.498
Peer Problems	3.855	2, 104	.024*
Social Acceptance	.062	2, 91	.940
Global Self-Worth	.181	2, 95	.835
Parent-Rated			
Total Difficulties	4.056	2, 108	.020*
Emotional Symptoms	7.470	2, 108	.007**
Peer Problems	5.275	2, 108	.940

Summary of Within-Group Analyses of Socio-Emotional Functioning

There was minimal support for gender and school stage differentiating disfigured young people on socio-emotional functioning with only one variable being significantly differentiated by age which was self-rated peer problems (with the primary school group reporting more peer problems than the secondary school group) and only one variable significantly differentiated by gender which was parent-reported Total Difficulties with boys rated worse than girls.

The medical variables were much more likely to differentiate socio-emotional functioning scores than demographic variables and, indeed, the presence of

disfigurement. Self-rated peer problems were significantly worse in participants with mobility problems, pain and a life threatening medical condition. On the medical variables with two categories, all but one parent-reported measure was significantly differentiated. Pain was the most consistent variable significantly differentiating all indices of socio-emotional measures except for self-reported Global Self-Worth. The multi-category medical variables indicated that participants with eczema and overgrowth syndromes did worse while those with vascular tumours were most likely to report the best socio-emotional functioning. Participants with the largest areas of their body affected by the disfigurement reported significantly worse functioning than those with less of their bodies affected, regardless of location of the disfigurement. On both disfigurement location and dermatological diagnosis analyses, the differences were found in nearly every parent-reported measure and in self-reported peer problems.

3.3 Aim 2: Attachment Security

The second aim of the current study was to compare the group of young people with congenital dermatological disfigurement against the general population on attachment security. The attachment measure used was the Child Attachment Interview (CAI)

Aim 2 tested the hypothesis that children and adolescents with congenital dermatological disfigurement would be significantly more likely to be insecurely attached then children and adolescents in the general population.

3.3.1 Analytic strategy

Frequency of attachment security and insecurity in the disfigured group was compared against published UK norms for the CAI (Schmueli Goetz et al., 2008) using a chi-square analysis.

The initial comparative analysis between the disfigured group and published UK norms was conducted using a three-way attachment classification; secure, insecure-dismissing and insecure pre-occupied for attachment to mother and attachment to father or a secondary attachment figure as well as a two-way attachment classification; secure and insecure. The subsequent analyses were conducted using the two-way attachment categorisation because the study hypotheses only distinguished between secure and insecure attachment categories rather than the specific insecure classifications individually. Attachment research commonly analyses data based on the two-way classification because the main theoretical differentiation occurs between the secure and insecure categories rather than between each insecure classification. Furthermore, each insecure classification tends to have far fewer group members than the secure group, which can pose problems for parametric statistical analyses and in terms of statistical power.

For the purposes of the current study, a single two-way classification of attachment consisting of a secure and insecure classification was constructed by combining attachment to mother and attachment to father. First, the two insecure categories of insecure dismissing and insecure pre-occupied attachment were combined. The single case of disorganised attachment was dropped from further analyses in order to meet the assumption of no fewer than five cases per cell for Chi-square analyses. Other measures for the single disorganised case also acted as outliers, which was a further justification for dropping the case from further analysis. Then, attachment to mother and attachment to father were compared and combined. In only 3 cases were attachment styles different between father and mother. In these cases, if either of the classifications was insecure, then the case was classified as insecure. In a further 3 cases, there was no attachment to father classification because the participant was unable to name a second attachment figure.

Attachment style was compared across all medical and demographic background variables to check for systematic within-group differences using the single two-way classification of attachment security. Group comparisons could not be made using a three-way attachment categorization as there were too few participants in the insecure categories to use non-parametric analysis. Only one significant difference was found on the variable of concomitant mobility problems with participants with mobility problems having a higher than expected chance of being insecurely attached (χ^2=7.52, p<.01). There were no other significant differences across attachment categories. The results of these analyses are shown in Appendix AA.

3.3.2 Findings

To test the hypothesis that children with a disfigurement may be at increased risk of an insecure attachment compared to the general population, a Goodness-of-fit Chi-Square Test for unequal expected values was used to compare the distribution of attachment classifications in the current sample against a non-clinical population sample published by the authors of the CAI. The attachment classifications for the disfigurement group are shown in Table 3.18. Table 3.19 shows the attachment classifications of the comparison group of 161 non-referred British children between 7 and 12 years of age (Shmueli-Goetz et al., 2008).

Table 3.18
Three Way Distribution of Attachment Classifications in the current sample

		Secure	Insecure		
	N	n (%)	Dismissing n (%)	Pre-occupied n (%)	Disorganised n (%)
Mother	100	58 (58)	32 (32)	9 (9)	1 (1.0)
Father	97	55 (56.7)	34 (35.1)	7 (7.2)	1 (1.0)

Table 3.19
Published Three Way Distribution of Attachment Classifications in a non-referred population (Schmueli-Goetz et al., 2008)

		Secure	Insecure		
	N	n (%)	Dismissing n (%)	Preoccupied n (%)	Disorganised n (%)
Mother	168	106 (66)	45 (28)	5 (3)	6 (4)
Father	154	97 (63)	43 (28)	8 (5)	6 (4)

There was a significant association between 3-way attachment classification to mother and dermatological disfigurement $\chi^2(2)= 13.23$, $p=.001$. The proportionate residual values suggested that the biggest difference between the two populations was that there were more insecure-pre-occupied participants in the current sample than in the normative sample. Cell chi-squared analyses confirmed that children with dermatological disfigurement were more likely than expected to have insecure preoccupied attachment style $\chi^2 (2) = 23.21$, $p=.001$.

The three-way classification for attachment to father showed no significant difference between the disfigured sample and the non-clinical sample $\chi^2(2)=3.01$, NS.

Table 3.20
Two-way Attachment Classifications in the Disfigured Group and the Published Norms.

	Secure n (%)		Insecure n (%)	
	Disfigured	General	Disfigured	General
Mother	58 (58)	106 (66)	42 (42)	56 (35)
Father	55 (56.7)	97 (63)	42 (43.3)	57 (37)

A further chi-square test on the two-way combined classification showed that there was a significant association between two-way attachment classification between the current sample and the normative data ($\chi^2(1)=4.76$, $p<.05$). This outcome represents the finding that, based on the odds ratio, the disfigurement group were 1.56 times more likely to be insecurely attached than the general population group.

The results supported the hypothesis that children and young people with congenital dermatological disfigurement are more likely to be insecurely attached than the normal population using a two-way attachment model and the three-way attachment to mother classification. The difference in attachment to father did not reach statistical significance between disfigured and general population groups despite a similar pattern of distribution to attachment to mother.

3.4 Aim 3 Shame

External shame, measured by the OAS (Goss et al., 1994b), and internal shame, measured by the ISS (Cook, 1996) were compared between the disfigurement group and a school-based comparison group.

Aim 3 tested the hypothesis that children and adolescents with congenital dermatological disfigurement would report significantly higher levels of external and internal shame compared to their peers in the general population.

3.4.1 Analytic strategy

An independent samples t-test was used to compare mean shame scores for the disfigurement group against the mean shame scores for a school-based comparison group.

Because there were no published norms for the age group investigated in the current study, comparison data for the two shame measures were collected from a school-based population. The procedure for the comparison group data collection is described in Appendix E. The sample characteristics for the comparison group are described in Table E.1 (Appendix E).

In order to determine if shame and general psychological functioning were associated in a similar way in the disfigured and non-disfigured populations, the association between each shame measure and the child-reported Total Difficulties subscale score, as an index of general mental health, was analysed in the disfigurement group and the non-disfigured comparison group. The correlations from each group were then compared using a Fisher's Z Test to determine if the relationship between shame and general mental health was similar or different in each group.

A Levene's test of variance was conducted to determine if there was a significant difference in variance of scores on each shame measure between the disfigurement and comparison groups.

A Pearson's Chi Square Analysis demonstrated that the disfigurement and comparison groups were not significantly different in gender representation. However, there were significantly more primary school aged children in the comparison group than in the disfigurement group χ^2 (2) = 6.79, p=.033. Statistical analyses were not used to compare ethnicity and occupational status between groups as the data used to describe the comparison group were local community estimates and not data specifically describing the group members. However, these local community estimates suggested that the disfigured group may have included substantially more white British participants (73.8%) than the comparison group (45.5%). There were no other marked differences in ethnicity and occupational category.

An independent samples two-tailed t-test was conducted to compare the disfigurement and comparison groups on the self-reported SDQ Total Difficulties subscale in order to compare general psychological functioning in each group. The

results indicated that the comparison group was significantly worse off $t(1,226)= 9.88$, $p<.001$.

External and internal shame scores reported by the disfigurement group were compared across key demographic, medical and disfigurement indices to identify any systematic within-group differences. There were no significant differences across demographic variables (gender, school stage, ethnicity and socio economic status). Across medical variables, there were no significant differences in shame based on age of onset, co-morbid medical problems, life threat, frequency of hospital attendance or diagnosis. There was a significant difference (t(df)=2.05, p=.04) between participants reporting pain and participants without pain with the former reporting higher levels of shame. Data of within group differences analyses according to demographic and medical variables are shown in Appendix BB.

3.4.2 Findings

To test the hypothesis that children with a disfigurement may be at increased risk of internal and external shame compared to the general population, a two-tailed independent samples t-test was used to compare the disfigurement group and comparison group mean scores on each shame measure. The results are shown in Table 3.21.

Table 3.21
Internal and External Shame in the Disfigurement and Comparison Groups

	Disfigurement		Comparison	
	N	M (SD)	N	M (SD)
Internal	87	20.7 (16.8)	158	31.42 (16.86)***
External	92	31.5 (11.3)	160	38.03 (11.03)***

The hypothesis that young people with congenital dermatological disfigurement would report higher levels of shame than the non-disfigured group was not supported by the current findings. In fact, the contrary was true, with the non-

disfigured comparison group reporting significantly higher internal and external shame. However, it should be noted that the non-disfigured comparison group also scored significantly higher on the measure of overall psychological functioning. This may indicate that the non-disfigured group had been experiencing a much higher level of psychological difficulty generally, which may have accounted for their higher levels of shame also.

The Levene's test of variance found that variance in internal and external shames scores in the disfigurement and comparison groups was not significantly different for either internal or external shame.

The association between each shame measure and child-reported Total Difficulties in each group was analysed using a Pearson's Two-Tailed Correlation Analysis. The bivariate correlations were then compared using a Fisher's Z Test. The correlation values between internal and external shame and the Total Difficulties scores for each group are displayed in Table 3.22. The two-tailed Fisher's Z scores are also shown for each correlation on the same table.

Table 3.22
Comparison of Correlations between Shame and General Psychological Difficulties

		Shame	
		Internal	External
Total Difficulties	Disfigured	.629**	.606**
	Non-disfigured	.550**	.501**
	Fisher's Z	.24	1.05

Internal Shame was significantly correlated with Total Difficulties for the disfigured group, r (110) = .629, p < .001, and for the non-disfigured group, r (115) = .550, p<.001. The difference between these correlations was not statistically significant, Z = .24, p=0.8103.

External shame was significantly correlated with Total Difficulties for the disfigured group, r (88) = .606, p< .001, and for the non-disfigured group, r(113) = .501, p<.001. The difference between these correlations was not statistically significant, Z = 1.05, p=0.294.

Shame and general psychological functioning were significantly correlated in both groups sampled. Results indicated that the relationship between these two variables was not significantly dissimilar in disfigured and non-disfigured young people.

3.5 Aim 4: Attachment, Shame and Socio-Emotional Functioning

Aim four consisted of three steps. The first step determined the association between each independent variable and the dependent variables. The second step determined if there was an association between the two independent variables. The third step determined if attachment and shame together were significantly associated with the measures of socio-emotional functioning.

Aim 4 tested the hypothesis that attachment and shame would together account for a significant amount of variance in socio-emotional functioning in children and young people with congenital dermatological disfigurement.

3.5.1 Step 1: Association between each independent variable and the dependent variables

3.5.1.1 Attachment and socio-emotional functioning

The aim of Step 1 was to determine if attachment and shame were independently associated with the measures of socio-emotional functioning. The first stage of Step 1 tested the hypothesis that socio-emotional functioning would be significantly better in the securely attached group compared to the insecure group.

3.5.1.1.1 Analytic Strategy

Each of the eight measures of socio-emotional functioning were compared across two-way attachment classifications using an independent samples t-test.

All outcome measures except for the Social Acceptance subscale (F=4.170, *p*=.044) where the insecure group showed a greater range of scores than the secure group met variance assumptions (Levene's Test for Equality of Variances). As such, the data were not transformed.

3.5.1.1.2 Findings

The mean scores on the socio-emotional functioning measures are shown below by attachment classification and comparison scores are shown in Table 3.23.

Table 3.23
Attachment and Socio-Emotional Functioning

	Secure	Insecure			
Self-report	M (SD)	M (SD)	*t*	df	*p*
Total Difficulties	8.62 (5.17)	13.29 (6.24)	3.91	89	.000***
Emotional Symptoms	2.56 (2.12)	3.56 (2.51)	2.06	89	.042*
Peer Problems	1.54 (1.66)	2.32 (1.93)	2.07	89	.042*
Social Acceptance	3.32 (.47)	3.13 (.60)	-1.66	83	.100
Global Self-Worth	3.26(.68)	3.00 (.56)	-1.87	84	.065
Parent Report					
Total Difficulties	8.41 (5.48)	9.49 (6.48)	0.89	95	.376
Emotional Symptoms	2.83 (2.54)	2.86 (2.65)	0.05	95	.959
Peer Problems	1.44 (1.72)	1.86 (1.95)	1.12	95	.268

**p<0.05; **p<0.01; *** p<0.001*

The secure attachment group reported significantly better scores than the insecure group on self-reported Total Difficulties, Emotional Symptoms and Peer Relationship Problems. There were no significant differences between secure and insecure groups on parent reported measures. Nor were there significant differences between attachment groups on the self-reported positive measures of Social

Acceptance and Global Self-Worth. However, there was a consistent trend for the secure group to show better functioning on all measures than the insecure group, albeit not reaching statistical significance.

The hypothesis that attachment security would significantly differentiate socio-emotional functioning was partially supported. Three of the five self-reported measure were significantly different across two-way attachment classification. Parent–reported measures and self-reported positive measures showed a non-significant trend to be better in the secure group than in the insecure group.

3.5.1.2 Shame and socio-emotional functioning.

The second stage of Step 1 was to determine if internal and external shame were associated with measures of socio-emotional functioning. The hypothesis tested was that internal and external shame would be significantly associated with socio-emotional difficulties.

3.5.1.2.1 Analytic strategy

A series of two-tailed, bivariate, Pearson's Correlation Analyses were conducted to investigate the association between shame and the measures of socio-emotional functioning. On both shame measures, the distribution of scores satisfied Levene's Test for Equality of Variance.

3.5.1.2.2 Findings

The correlations between internal and external shame and socio-emotional functioning are presented in Table 3.24.

Table 3.24
Shame and Socio-Emotional Functioning

	External Shame	Internal Shame
Self-Reported		
Total Difficulties	.606***	.629***
Emotional Symptoms	.507***	.589***
Peer Problems	.517***	.506***
Social Acceptance	-.565***	-.454***
Global Self-Worth	-.613***	-.671***
Parent-Reported		
Total Difficulties	.276**	.360**
Emotional Symptoms	.181	.308**
Peer Problems	.317**	.390***

**p<0.05; **p<0.01; *** p<0.001*

All self-reported measures of socio-emotional functioning were significantly correlated with internal and external shame. Global Self-Worth showed the biggest correlation with internal shame and external shame with internal shame accounting for 45% of the variance of Global Self-Worth and external shame accounting for 37.6% of the variance in the Global Self-Worth score. Internal shame accounted for 39.6% of the variance in the self-reported Total Difficulties score.

The relationships between internal shame and external shame and the parent reported indices of socio-emotional functioning were slightly weaker. Parent ratings of Peer Problems showed the strongest correlation with internal and external shame. Parent-rated Total Difficulties was also significantly related to both internal and external shame. Parent-rated Emotional Symptoms was significantly associated with internal shame but not with external shame.

In conclusion, there was support for the hypothesis that shame would be significantly associated with socio-emotional functioning. The relationship between both shame measures and socio-emotional functioning measures appeared to be

stronger on the self-reported measures although still highly correlated with all but one parent-reported measure.

3.5.2 Step 2: Attachment and Shame

Step 2 aimed to investigate the relationship between attachment and shame. It tested the hypothesis that mean shame scores would be significantly different across attachment security categories with the insecure group reporting higher internal and external shame.

3.5.2.1 Analytic Strategy

The relationship between attachment and shame was assessed by comparing mean shame scores across attachment categories. An independent samples t-test was used to evaluate differences on shame scores between the combined two-way attachment categorization.

3.5.2.2 Findings

The result of the independent samples t-test showed that there was no significant difference between secure and insecure attachment classifications on external shame but that there was a significant difference on internal shame ($t(73) = 2.85$, $p = .006$) as shown in Table 3.25. Figure 3.1 below demonstrates that there was a trend towards participants with secure attachment style having lower internal and external shame than participants with insecure attachment style, despite the latter difference not reaching statistical significance.

Table 3.25
Shame and Attachment

	Secure (n=43)	Insecure (n=32)
Internal	17.33(13.49)*	29.25(22.59)
External	30.79(11.39)	34.75(11.37)

**p<0.05; **p<0.01; *** p<0.001*

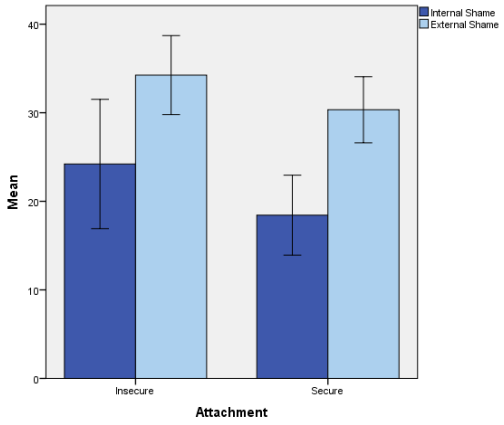

Figure 3.1 Shame and Attachment (95% CI)

These results support the hypothesised relationship between attachment and shame. That is, that there would be a weak but significant association between the two variables.

3.5.3 Step 3: Attachment, shame and psychosocial functioning.

The aim of Step 3 was to test the hypothesis that attachment and shame will together account for a significant amount of variance in socio-emotional functioning.

3.5.3.1 Analytic Strategy

A series of hierarchical regression analyses were conducted to test the association between attachment and shame on measures of socio-emotional functioning in children and adolescents with disfiguring dermatological conditions. Regression analyses were based on a two-way secure/insecure attachment classification. Attachment was entered first, followed by shame as per the hypothesized causal pathway.

In both the disfigurement and comparison groups, OAS and ISS scores were found to be highly correlated using a Pearson's bivariate, two-tailed correlational analysis ($r(90)=.83$, $p<.001$) and ($r(153)=.81$, $p<.01$) respectively. A correlation of more than .80 is typically considered so high as to suggest that the two measures are accessing the same underlying construct (Taylor, 1990). As such, it was decided that only one measure of shame should be included in the remaining analyses. External shame (measured by the OAS) was retained, rather than internal shame, for conceptual reasons. External shame is central to the causal relationship proposed in the explanatory framework as it is defined as the emotional, cognitive and behavioural reaction to actual or perceived social rejection. Internal shame describes the phenomenon whereby perceived rejection by others becomes internalised by the individual so that they view themselves as rejected or "rejectable" also. Because internal shame is the second order process and because the internalization of external experiences and messages in children is still not clearly understood (Barenboim, 1981) internal shame was excluded from the remaining analyses.

Collinearity diagnostics were conducted on all analyses, which indicated no violations of linearity, normality or homoscedasticity.

3.5.3.2 Findings

In relation to the self-reported Total Difficulties subscale, the results indicated that the two independent variables, together, explained just under half of the variance ($R^2=.418$, $F(2,77)=27.63$, $p<.001$). Both independent variables were also, separately, significantly associated with the outcome variable. These data are shown in Table 3.26 below.

Table 3.26
Self-reported SDQ Total Difficulties

	B	*SE B*	*β*
Step 1			
Constant	13.171	.924	
Attachment	-5.216	1.233	-.432***
Step 2			
Constant	3.970	1.841	
Attachment	-3.938	1.075	-.326***
Shame	.258	.047	.492***

Note R^2 = .187 for Step 1; ΔR^2 = .231 for Step 2
*p<0.05; **p<0.01; *** p<0.001

In relation to the self-reported Emotional Symptoms subscale, attachment and shame together explained nearly one quarter of the variance (R^2=.234, F(2,77)=11.77, p<.001). Both independent variables were, individually, significantly associated with the outcome variable; attachment (β = -.238, p<.05) and shame (β = .431, p<.001) as shown in Table 3.27.

Table 3.27
Self-Reported SDQ Emotional Symptoms

	B	*SE B*	*β*
Step 1			
Constant	3.429	.379	
Attachment	-1.095	.506	-.238*
Step 2			
Constant	.357	.804	
Attachment	-.669	.469	-.145
Shame	.086	.020	.431***

Note R^2 = .057 for Step 1; ΔR^2 = .177 for Step 2
**p<0.05; **p<0.01; *** p<0.001*

In relation to the self-reported Peer Problems subscale, the independent variables together explained a significant portion of the variance (R^2=.161, F(2,77)=7.41, p=.001). Attachment was not independently significantly associated with peer problems (β = -.200, p>.05) but shame was significantly associated with self-reported peer problems (β = .357, p=.001). The data are shown in Table 3.28.

Table 3.28
Self-Reported SDQ Peer Problems

	B	*SE B*	*β*
Step 1			
Constant	2.171	.293	
Attachment	-.705	.391	-.200
Step 2			
Constant	.227	.644	
Attachment	-.435	.376	-.124
Shame	.054	.016	.357***

Note R^2 = .040 for Step 1; Δ R^2 = .121 for Step 2
**p<0.05; **p<0.01; *** p<0.001*

In relation to self-reported social acceptance, attachment and shame, together, explained a significant portion of the variance (R^2=.164, F(2,77)=7.14, *p*=.001). Each attachment (*β* = .248, *p*<.05) and shame (*β* = -.326, *p*<.01) accounted for a significant amount of variance in social acceptance independently of one another as shown on Table 3.29.

Table 3.29
Self-Reported SPP Social Acceptance

	B	*SE B*	*β*
Step 1			
Constant	3.084	.093	
Attachment	.268	.122	.248*
Step 2			
Constant	3.638	.205	
Attachment	.197	.118	.182
Shame	-.015	.005	-.326**

Note R^2 = .061 for Step 1; Δ R^2 = .102 for Step 2
**p<0.05; **p<0.01; *** p<0.001*

In relation to self-reported Global Self-Worth, attachment and shame, together, explained a significant amount of the variance (R^2=.295, F(2,77)=15.49, *p*<.001). Both attachment (*β* = .171, *p*>.05) and shame (*β* = -.481, *p*<.001) were independently significantly associated with global self-worth as shown in Table 3.30.

Table 3.30
SPP Global Self-Worth

	B	*SE B*	*β*
Step 1			
Constant	2.957	.111	
Attachment	.354	.145	.272*
Step 2			
Constant	3.938	.226	
Attachment	.222	.130	.171
Shame	-.027	.006	-.481***

Note R^2 = .074 for Step 1; Δ R^2 = .221 for Step 2
**p<0.05; **p<0.01; *** p<0.001*

In relation to parent-reported Total Difficulties, the results of the regression indicated that the two independent variables, together explained a small amount of the variance (R^2=.040, F(2,77)=1.65, p>.05) which was not significant although each attachment and shame were independently, significantly associated with the outcome measure. These data are shown in Table 3.31.

Table 3.31
Parent-Report SDQ Total Difficulties

	B	*SE B*	*β*
Step 1			
Constant	10.704	1.041	
Attachment	-3.114	1.354	-.276*
Step 2			
Constant	8.212	1.390	
Attachment	-2.581	1.314	-.229
Shame	.103	.040	.300*

Note R^2 = .021 for Step 1; Δ R^2 = .019 for Step 2
**p<0.05; **p<0.01; *** p<0.001*

In relation to parent-reported Emotional Symptoms, the two independent variables, together, explained little variance (R^2=.014, F(2,77)=.560, p>.05) which was not significant. Each individual independent variable did not account for a significant portion of the variance in the parent-reported Emotional Symptoms score. The data are shown in Table 3.32.

Table 3.32
Parent-Reported SDQ Emotional Symptoms

	B	*SE B*	*β*
Step 1			
Constant	2.743	.406	
Attachment	-.126	.536	-.026
Step 2			
Constant	1.864	.944	
Attachment	-.025	.545	-.005
Shame	.025	.024	.117

Note R^2 = .001 for Step 1; ΔR^2 = .013 for Step 2
**p<0.05; **p<0.01; *** p<0.001*

In relation to parent-reported peer problems, attachment and shame, together, explained a small, non-significant portion of the variance (R^2=.063, F(2,77)=2.649, *p*>.05). These data are shown in Table 3.33.

Table 3.33
Parent-Reported SDQ Peer Problems

	B	*SE B*	*β*
Step 1			
Constant	1.971	.299	
Attachment	-.610	.395	-.170
Step 2			
Constant	.925	.686	
Attachment	-.490	.396	-.137
Shame	.029	.017	.187

Note R^2 = .029 for Step 1; ΔR^2 = .034 for Step 2
**p<0.05; **p<0.01; *** p<0.001*

Overall there was support for the hypothesis that attachment and shame, together, would account for a significant amount of variance in socio-emotional functioning. Attachment and shame, together, accounted for significant amounts of variance in all self-reported outcome measures. Variance on the parent-reported measures was not significantly explained by attachment and shame. Change scores indicated that attachment and shame accounted for independent and overlapping variance in outcome measures.

4.0

POST HOC STUDY:

DISFIGUREMENT VISIBILITY AND PSYCHOLOGICAL FUNCTIONING

4.1 Introduction

While existing research has not supported the influence of the disfigurement related variables of severity or disease-origin, there are findings to suggest that the visibility of the disfigurement may play a more significant role. At the start of the current project there were only limited findings supporting the influence of visibility with no published studies unequivocally supporting this opinion which was shared in two position papers by Rumsey & Harcourt (2004) and Thompson & Kent (2001). After the current study was underway, findings of the large-scale ARC study (Clarke et al, 2013) reported significant findings to support the influence of visibility of disfigurement on psychosocial functioning. The study found that self-rated visibility of the disfigurement significantly distinguished scores on social anxiety, general anxiety, depression, aggression, negative affect and positive affect with the group reporting visible disfigurement reporting significantly worse function on these measures.

In contrast, a study on children with cleft lip and palate (CLP) (Feragen et al, 2010) reported that children with a visible cleft reported significantly better scores on self-perceived social acceptance and close friendships as measured by the Harter Self-Perception Profile (Harter, 1985) and better depression scores, as measured by the Hopkins Symptom Checklist (Dean, Leathern & Spicer, 1992) than the general population sample. The same study also reported no significant differences between the visible cleft group and the non-visible cleft group on measures of close friendships, social acceptance and depressive symptoms. However, they did find significantly lower appearance satisfaction in girls with a visible cleft than girls with a non-visible cleft. This difference was not replicated in boys with CLP.

The findings of the Clarke group are important as the first robust findings to indicate that a disfigurement-related variable might significantly influence psychological

functioning. These findings are also consistent with the notion, on which the main study was based, that being viewed as disfigured by others might play a key role in influencing negative psychological impact on the disfigured person. However, the findings of the Clarke study are limited by the fact that they are based on adults only and that they are based on self-reported measures of visibility and psychological functioning, hence potentially reflecting shared-method variance rather than a true association between variables. Furthermore, the relationship between visibility of disfigurement and psychological adjustment has not been found in a recent study on adolescents with CLP.

The aim of this post hoc study was to attempt to replicate the recent findings of the largest study on the psychological impact of disfigurement to date – i.e. that the visibility of the disfigurement might influence psychological functioning. This study aimed to test the Clarke findings on a child and adolescent population rather than an adult population. This study also used a measure of disfigurement visibility that was reported by the child participant's parent which would test the possibility that the findings of the Clarke study might have been the result of shared method variance rather than a true association between disfigurement visibility and psychological functioning.

4.2 Method

4.2.1 Design

The study set out to test the hypothesis that disfigurement visibility would differentiate psychological functioning with those with a visible disfigurement reporting more psychological difficulties than those with a non-visible disfigurement. Parent-reported data on the location of the disfigurement were used to group the participants into a visible disfigurement and a non-visible disfigurement group. All psychological measures collected in the current study were then compared between

the two groups in order to determine if visibility of disfigurement significantly differentiated psychological functioning.

4.2.2 Measures

4.2.2.1 Visibility of Disfigurement

As part of the main study, parents were asked to provide information about the location of their child's disfigurement. This information was collected in two ways. First, parents were given two line drawings that were exactly the same that represented the front of their child and the back of their child. The parent was then asked to shade the areas of their child's body that were currently or typically affected by their disfigurement. Second, in order to form a categorical index of disfigurement location, the researcher coded the disfigurement location into seven categories: face only, limbs only, torso only, face and limbs, face and torso, limbs and torso and complete body.

For the purposes of the post hoc study, disfigurement that was located on the face, neck and lower arms and hands were considered to be visible. These were the areas of the body considered to be typically "visible" to others. Legs and feet were not considered to be visible as they are easily and often concealed by trousers, tights, socks and shoes.

In order to group each participant as having visible or non-visible disfigurement, the shaded diagrams of disfigurement location were reviewed by the researcher and classed as visible if the shaded areas included the face, neck, lower arms or hands. Remaining participants were classed as non-visible. The categorical data, based on the shaded diagrams, were then checked to ensure that the visible-non-visible classification was consistent with these data also.

4.2.2.2 Psychological Functioning

The same measures of psychological functioning were used in the follow-up study as in the primary study. The measures are listed in Table 4.1. Each measure is described in more detail in Section 2.3.

Table 4.1
Measures of Psychological Functioning

Measures	Section	Appendix
Self-Reported Measures		
SDQ Total Difficulties Emotional Symptoms Peer Problems	2.3.2.1	Mi
SPP Social Acceptance Global Self Worth	2.3.2.2	N
Other as Shamer Scale	2.3.3.2	R
Internalised Shame Scale	2.3.3.3	S
Parent-Reported Measures		
SDQ Total Difficulties Emotional Symptoms Peer Problems	2.3.2.1	Mii
Demographic Questionnaire	2.3.1.1	K
Medical Questionnaire	2.3.1.2	L
Independently Rated Measures		
Child Attachment Interview	2.3.3.1	O

4.2.3 Sample

The participants were drawn from the 122 participants aged between 8 and 16 years included in the main study. The sampling procedure is described in Section 2.4.2 and the sampling selection, including inclusions and exclusion criteria, is described in Section 2.2. Participants who had missing disfigurement location data were excluded from the study. As such, the total number of participants included in the post hoc study was 114.

4.2.4 Procedure

- Participants of the main study were assessed using the disfigurement visibility protocol described in Section 4.2.2.1
- Two groups: a visible group and a non-visible group were created.
- The visible and non-visible groups were compared on all psychological measures used in the main study.

4.3 Results

4.3.1 Sample Characteristics

Of the 114 participants included in the post hoc study, 78 were classed as having a visible disfigurement and 36 were classed as having a non-visible disfigurement.

Table 4.2
Disfigurement Locations and Groupings

Location	n (%)	Visible
Face only	33 (27.7)	
Hands/Lower Arms	9 (7.6)	78
Face and Hands	4 (3.4)	
Whole Body	26 (21.8)	
Face and torso	6(5.0)	
		Non-Visible
Torso only	9 (7.6)	
Limbs and torso	8 (6.7)	36
Legs/Feet/Upper Arms	19 (15.9)	
Missing	5 (4.2)	
N		**114**

The visible and non-visible disfigurement groups were compared on all demographic and medical variables. The sample characteristics for each group are displayed in Table 4.3.

Table 4.3
Demographics

	Visible	Non-Visible
Age/School		
Mean Age	144m (12yrs, 0m)	151m (12yrs, 6m)
Age Range	8yrs 0m - 16yrs 11m	8yrs 4m – 16yrs 11m
Primary School	33 (42.3%)	15 (41.7%)
Secondary School	44(56.4%)	21 (58.3%)
Gender	n (%)	
Female	46 (59)	23 (63.9)
Male	32 (41)	13 (36.1)
Ethnicity Category	n (%)	
White	68 (87.2)	32 (88.9)
Mixed Race	5 (6.4)	1 (2.8)
Asian	2 (2.6)	2 (5.6)
Black	2 (2.6)	0 (0.0)
Other (inc. Arab)	1 (1.3)	0 (0.0)
Unknown	0 (0)	1 (2.8)
Occupational Category	n (%)	
Professional	30 (38.4)	9 (25.7)
Higher Management	20 (25.6)	10 (27.8)
Technical/Craft	8 (10.3)	4 (11.1)
Lower Supervisory	6 (7.7)	5 (13.9)
Semi routine	3 (3.8)	1 (2.8)
Intermediate	2 (2.6)	2 (5.6)
Routine	2 (2.6)	2 (5.6)
Unemployed (inc benefits)	5 (6.4)	2 (5.6)
Unclassified	2 (2.6)	1 (2.8)

Grouped Condition	n (%)	n(%)
Vascular Birthmarks	29 (37.2)	19 (52.8)
Eczema	14 (17.9)	5 (13.9)
Bullous Disorder	12 (15.4)	1 (2.8)
Overgrowth Syndrome	4 (5.1)	4 (11.1)
Naevii	7 (9.0)	4 (11.1)
Other	7 (9.0)	1 (2.8)
Mastocytoses	5 (6.4)	2 (5.6)
Age of Onset	n(%)	
Birth	60 (76.9)	31 (86.1)
Birth - 24 mths	18 (23.1)	5 (13.9)
Hospital Visits	n (%)	
0-2	50 (64.1)	26 (69.5)
3-5	16 (20.5)	7 (19.4)
6 or more	9 (11.5)	2 (5.6)
Missing	3 (3.8)	1 (2.8)

Table 4.4
Other Medical Features

	Yes n (%)		No n (%)		Unknown n (%)	
	Visible	Non-	Visible	Non-	Visible	Non-
Pain	25 (32.1)	5 (13.9)	53 (67.9)	31 (86.1)	0 (0.0)	0 (0.0)
Mobility problems	29 (37.2)	7 (19.4)	49 (62.3)	28 (77.8)	0 (0.0)	1 (2.8)
Other problems	27 (34.6)	5 (13.9)	51 (65.4)	31 (86.1)	0 (0.0)	0 (0.0)
Potentially fatal	16 (20.5)	5 (13.9)	57 (73.1)	29 (80.6)	5 (6.4)	2 (5.6)

A chi-square test of independence found that there were no significant differences in frequencies of each variable across the visible and non-visible groups. Ethnicity and socio-economic status were not compared using inferential statistical analysis, as

the sub-group numbers did not reach the minimum required. However, on face value, the number of white versus non-white participants in the visible (87.2%) and the non-visible (88.9%) groups was very similar. However, the percentage of participants whose main earning parent was categorized as professional or higher management in the visible group (64.0%) seemed somewhat higher than in the non-visible group (53.5%).

4.3.2 Findings

An independent samples t-test was used to compare mean scores on all continuous measures of psychosocial functioning. Levene's Test for Equality of Variances found that variance of scores in each group were not significantly different. The results of the t-test are summarized in Table 4.5.

Table 4.5
Psychological Functioning in Visible and Non-Visible Disfigurement

Child Reported	Visible X(SD) n=78	Non-Visible X(SD) n=36	t	df	*p*
Total Difficulties	10.51 (6.07)	10.41 (6.00)	.084	104	.933
Emotional Symptoms	2.93 (2.47)	2.84 (1.74)	.184	82.06	.834
Peer Problems	1.88 (2.00)	1.78 (1.64)	.241	104	.810
Social Acceptance	3.21 (.58)	3.27 (.52)	-.429	93	.669
Global Self Worth	3.20 (.62)	3.05 (.65)	1.169	98	.245
Internal Shame	20.55 (15.48)	22.19 (20.14)	-.407	80	.685
External Shame	31.68 (11.50)	32.08 (11.56)	-.148	85	.883

Parent Reported	Visible X(SD)	Non-Visible X(SD)	t	df	*p*
Total Difficulties	9.01(6.01)	7.63 (5.64)	1.119	108	.266
Emotional Symptoms	2.72 (2.42)	2.78 (2.59)	-.122	108	.903
Peer Problems	1.77 (1.94)	1.28 (1.61)	1.255	108	.212

A chi-squared analysis of two-way attachment by visibility of disfigurement indicated that there was no significant skew in the distribution of attachment categories across the two disfigurement visibility groups (χ^2 (1) = 2.223, p >.05). However, it was notable that more participants with non-visible disfigurement were insecurely attached than securely attached. In the visible disfigurement group, as in the

general population, approximately 60% of the population is securely attached. In the non-visible disfigurement group, the percentages are inversed. Distribution of attachment security across visibility groups is shown in Table 4.6.

Table 4.6
Attachment in Visible and Non-Visible Disfigurement

	Visible (%)	Non-visible (%)
Secure	40 (58.8)	12 (42.8)
Insecure	28 (41.2)	16 (57.2)
Total	68 (100)	28(100)

4.3.3 Summary of Findings

The aim of the post hoc study was to test if visibility of disfigurement differentiated socio-emotional functioning in the sample of 8-16 year olds with congenital dermatological disfigurement. The hypothesis, based on the findings of the adult study by Clarke et al (2013), was that young people with a visible disfigurement would report greater psychosocial difficulties than the young people with a non-visible disfigurement.

The results did not support the hypothesis that young people with visible disfigurement would report worse psychosocial difficulties than young people with a non-visible disfigurement. Not one of the measures used in this study was significantly differentiated by disfigurement visibility. These findings are in direct contrast to the findings of the ARC study (Clarke et al., 2013). However, they are consistent with the findings of Feragen et al. (2010) who reported no significant differences between their visible and non-visible CLP groups, except for appearance satisfaction being lower in girls in the visible cleft group.

There were some limitations to this study. First, the measurement of disfigurement visibility was based on disfigurement location data. While the collected information allowed a fairly reliable categorization to be made, in particular the pictorial

representation of disfigurement location completed by a parent, a planned means of grouping participants according to visibility of disfigurement might have been more reliable. Second the final groupings were uneven, therefore, potentially introducing some error into the statistical analyses conducted.

The findings of the post hoc study are considered in the wider context of the main study in the Discussion, Chapter Five. Key conceptual and methodological issues highlighted by the post hoc study are also considered in the main Discussion chapter.

5.0

DISCUSSION

5.1 Introduction

This chapter restates the aims of the current study, summarises the results and links the findings with the wider literature. Key conceptual, methodological and empirical issues that were highlighted by the findings of this study are discussed. Finally, some empirical limitations and implications for future research and clinical practice are discussed.

5.1.1 Purpose of the study.

While the psychology of appearance has been a major topic of empirical and socio-cultural interest for some time, the psychology of individuals with a disfigured appearance has been relatively, empirically neglected. The existing disfigurement literature is further constrained by its focus on individuals with acquired disfigurement such as burns, individuals with CLP and adults. Furthermore, many studies on disfigured populations have been descriptive. This is understandable since a coherent and consistent description of the psychological function of children with disfigurement is yet to be unequivocally determined. Fewer studies have explored the specific developmental pathways that might account for the psychosocial heterogeneity in this population reported by existing studies.

The current study aimed to develop the existing body of research on the socio-emotional functioning of people affected by disfigurement by addressing some of the sampling limitations of the current literature. That is, by measuring socio-emotional functioning in congenital dermatological disfigurement and not CLP and by focusing on children and adolescents rather than adults. The purpose of the study was, further, to try to identify key developmental factors that contribute to the broad range of socio-emotional functioning in this population drawing on the now substantial literature on psychological development to form a coherent conceptual framework.

This study tested an explanatory model of socio-emotional adjustment in children and teenagers with congenital dermatological disfigurement. Based on the developmental psychopathology framework, disfigurement was positioned as a psychological risk factor due to the negative stance towards people with disfigured appearance. These negative social interactions were seen as a chronic stressor. Two independent variables were included in the model that were considered to influence the way in which the disfigured individual might respond to social stress that might ultimately result in either positive or negative socio-emotional functioning. These factors were attachment and shame. The dependent variable of socio-emotional functioning was selected based on a systematic review of the existing literature on psychological adjustment in disfigured children and adolescents. This study hypothesized that children and adolescents with congenital dermatological disfigurement would be at risk of socio-emotional difficulties because of their experiences of social rejection relating to their appearance. The study further hypothesized that attachment and shame might interact with congenital disfigurement to significantly influence socio-emotional functioning. The explanatory model that was tested by the current study is represented in Figure 5.1. The aims and hypotheses of the study are listed in Table 5.1.

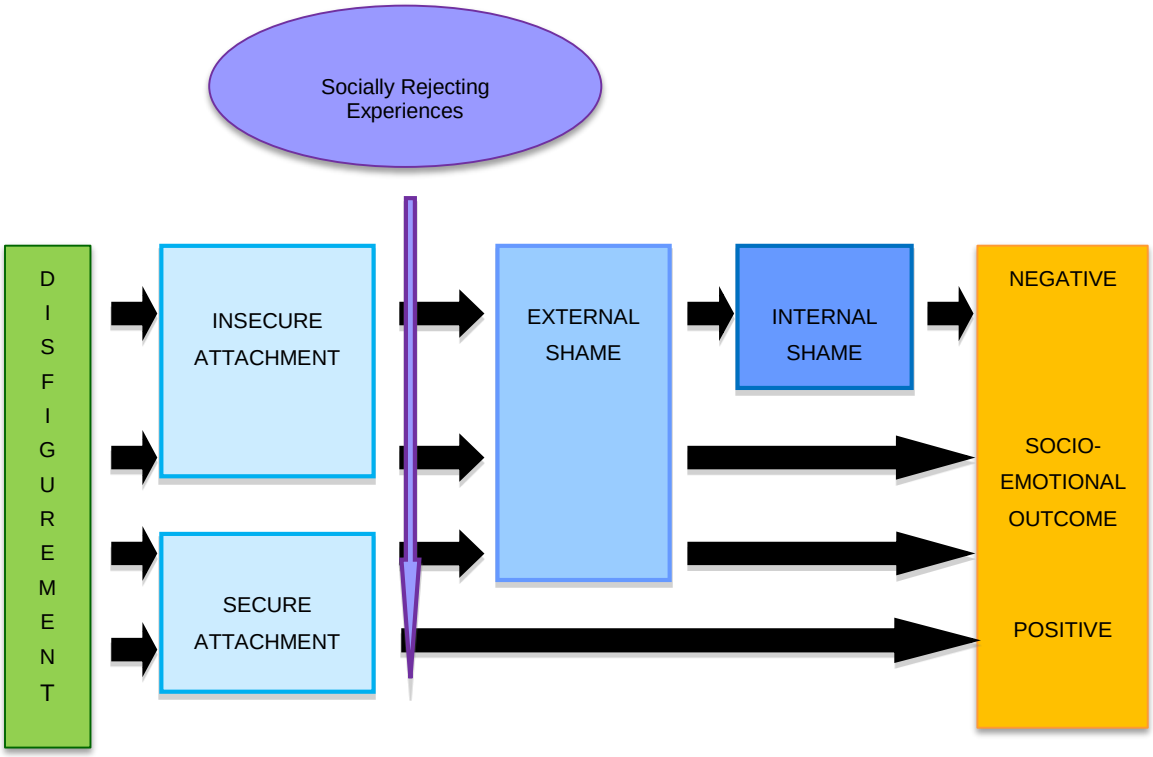

Figure 5.1. Explanatory Model of Socio-Emotional Functioning in Young People with Congenital Disfigurement

Table 5.1

Study Aims and Hypotheses

	AIMS	HYPOTHESES
1	To describe the socio-emotional profile of young people with congenital dermatological disfigurement	Young people with congenital dermatological disfigurement will have higher levels of socio-emotional difficulty than the general population
2	To measure attachment style in young people with congenital dermatological disfigurement.	Young people with congenital disfigurement will be more likely to be insecurely attached than the general population.
3	To assess internal and external shame in young people with congenital dermatological disfigurement	There will be higher internal and external shame in young people with disfigurement than the general population.
4	To assess the association between attachment and shame in relation to socio-emotional functioning.	Attachment and shame will be significantly associated, combined and independently, with socio-emotional functioning.

5.2 Summary of Results

5.2.1 Aim 1

The first aim of the study was to compare the disfigured population with the general population on measures of emotional adjustment and social relationships. The results revealed that, overall, the disfigured group were not significantly different to the population norms. The disfigured group scored significantly worse on parent-reported emotional symptoms and significantly better on self-reported global self-worth and social acceptance compared to population norms. The disfigured young people were more than twice as likely to fall within the “abnormal” classification on the parent-reported measure of emotional symptoms. The disfigured group was also significantly more likely to fall within the “borderline” clinical range for self-reported total difficulties than the general population, despite there being no significant difference between group means on this measure. All other indices of psychological functioning were not significantly different from the general population.

Overall, these findings did not unequivocally support the hypothesis that the disfigured group would report significantly greater social and emotional problems than the general population. These findings were, however, consistent with the existing literature in that there are no unequivocal findings of socio-emotional deficits in congenitally disfigured children and young people compared to the general population. These findings were also consistent with the notion of socio-emotional heterogeneity in the disfigured population in that there was evidence of increased rates of caseness on some measures despite group means that were not significantly different from population norms.

Within group analyses based on gender showed no significant differences except for one measure (parent-reported Total Difficulties where boys were scored as

significantly worse than girls) suggesting that gender did not have a significant impact on socio-emotional functioning in this sample. Similarly, the age proxy-variable of school stage only differentiated on one measure of socio-emotional functioning. Self-reported social acceptance was significantly lower in the primary school group. Overall, there was little support for these demographic variables of age and gender to have a main effect relationship with socio-emotional functioning in this population. This finding is in keeping with earlier studies that have reported largely no effect of demographic variables but where there have been weak effects reported for age and gender (Feragen et al, 2010; Leonard et al, 1991; Shute et al, 2007).

The findings relating to within group differences based on medical variables were more compelling. While specific disfigurement locations, such as the face, did not individually differentiate socio-emotional functioning scores, the pattern of significant differences indicated that participants with larger total areas of the body affected by the disfigurement reported significantly poorer functioning on three of the eight socio-emotional indices: self-reported peer problems, parent-reported total difficulties and parent-reported peer problems. The findings based on diagnostic groupings were similar with significant differences on the same three outcome measures plus parent-reported emotional symptoms also. Significantly worse functioning was reported in participants with eczema and overgrowth syndromes in comparison to participants in the vascular tumour group. These two sets of findings present a potentially consistent picture in that eczema and overgrowth syndromes are more likely to affect larger areas of the body while vascular tumours tend to be small, discrete lesions, albeit often very visible such as on the face. However, it is impossible to determine from these findings if area of body affected is the key, influencing factor, or if the diagnosis is more influential. The presence of pain, mobility problems and co-morbid medical problems all significantly differentiated the

majority of socio-emotional measures. These three variables each significantly differentiated all parent-reported measures and self-reported Total Difficulties and Peer Problems. Pain was the most consistent discriminating variable significantly differentiating all outcomes except for self-reported Global Self-Worth. The effects were the strongest for the parent-reported outcomes. The final medical variable of life threat also significantly differentiated self-reported peer problems and parent-reported Total Difficulties and Emotional Symptoms but not parent-reported Peer Problems.

In summary, while age and gender did not seem to distinguish social emotional functioning effectively, the medical factors were more successful in differentiating socio-emotional functioning scores. In fact, every medical factor tested was more likely to differentiate outcomes than the presence of disfigurement. The presence of pain, in particular, was associated with significantly worse socio-emotional functioning.

These findings are consistent with those reported by Feragen, Stock & Rumsey (2014) that medical co-morbidities distinguished their population of young people with CLP on indices of psychological functioning leading them to highlight the importance of building in medical co-morbidities and key medical symptoms into research on young people with disfiguring medical conditions. These findings also resonate with the wider literature on young people with dermatological conditions in that children with eczema have been more likely to be found to have significant psychological dififculties (e.g., Absolon et al, 1997) than young people with other dermatological conditions such as CMN (Koot et al., 2000) and haemangioma (Zweegers & van der Vleuten, 2012). These findings further challenge the ARC explanatory model of psychological functioning in disfigured adults (Clarke et al., 2013) which focuses on internal psychological processes rather than physical

aspects of the disfigurement. While these findings do not refute the role of internal processing, they suggest an important role for medical variables as well.

These findings suggest that further research on psychological functioning in young people with dermatological conditions might be more usefully focused on the effects of other signs and symptoms of these conditions as well as on disfigurement.

5.2.2 Aim 2

The second aim of this study was to test the hypothesis that the disfigured group would demonstrate a significantly increased rate of attachment insecurity compared with the population norm due to theorized barriers to optimal parent-infant interaction relating to congenital disfigurement. This hypothesis was largely supported by the results. Based on a two-way attachment classification, the disfigured group was significantly more likely to display insecure attachment style than the published norms. The comparison based on a three-way attachment classification indicated that the biggest significant difference in attachment categorization between the disfigured and the general population groups was in a greater number of the disfigured group in the insecure-pre-occupied attachment category.

While there have been no other studies published on attachment style in congenitally disfigured school-aged children, this finding is contrary to the findings of Speltz, Endriga, Fisher & Mason (1997a), Maris et al (2000) and Murray et al. (2008) all of whom concluded that there were no lasting differences in rates of attachment security between very young children with CLP and the general population. The findings of the current study are, however, consistent with the studies conducted by Rabung et al. (2004), Picardi et al. (2003b) and Picardi et al. (2005) who reported that their samples of adults with dermatological conditions were

significantly more likely to report attachment insecurity. The consistency of these findings in studies on middle childhood through to adult disfigured groups may be interpreted as challenging the infant studies that found no attachment differences. Alternately, increased attachment insecurity in the older groups may represent discontinuity in attachment stability over time. These findings may suggest that attachment style in disfigured people may be sensitive to other influences over time that increase the likelihood of attachment insecurity beyond infancy.

5.2.3 Aim 3.

The third aim of the study was to test the hypothesis that the disfigured group would report significantly higher levels of internal and external shame than the non-disfigured population, because of the higher levels of social rejection experienced by this group. The results indicated that the disfigured group reported significantly lower levels of internal and external shame than the comparison group, which did not support the hypothesis being tested. However, these results should be interpreted with caution as the comparison group also reported significantly worse overall psychological functioning, which may indicate that the comparison group was not representative of the general population. Rather, they were experiencing relatively high levels of psychological dysfunction at the time of participation. The relationship between shame and psychological functioning was similar in both groups showing a high correlation between each shame measure and general psychological functioning. These correlations were not significantly higher in the disfigurement group when analysed despite the co-efficients being higher for both shame measures and psychological functioning in the disfigurement group.

Only one previous, unpublished study compared shame between disfigured and non-disfigured young people (Mason et al., 2009). The results of the current study

are consistent with those of the Mason study in that the general population was found to report significantly higher levels of shame than the disfigured groups. These findings may provide some challenge to the assumption that either disfigured young people experience more social rejection than non-disfigured children or that disfigured children are affected in a predictable way by socially rejecting experiences. This is also consistent with the opinions shared in recent publications that there is currently no empirical evidence to support the notion that disfigured young people experience any more explicit social rejection than their non-disfigured peers (e.g., .Feragen & Borge, 2010; Lovegrove & Rumsey, 2005) but is in contrast to some adult studies that reported much higher rates of negative social experiences (e.g., Strauss et al., 2007).

The assumption of negative social experiences is central to the hypothesis that disfigured young people are at risk of psychological and social difficulties because of the impact of social rejection and bears further research.

5.2.4 Aim 4

The fourth and final aim of the study was to determine if attachment and shame, together, would account for a significant amount of variance in socio-emotional functioning in the disfigured group. The first step of this aim was to determine if each independent variable was significantly associated with the outcome variables. Attachment style was found to be significantly associated with all of the self-reported outcome measures while shame was found to be significantly associated with all outcome variables. Self reported outcome variables were more likely to be significantly associated with the independent variables or have higher amounts of variance explained by the independent variable.

The association between attachment style and shame was also evaluated. Using a two-way categorization of attachment security, only internal shame, and not external shame, was found to be significantly associated with attachment. However, there was a non-significant trend for external shame, as well, to be higher in the insecure group than in the secure group. Because both shame measures were highly correlated, only the external shame measure was used in the final explanatory model that was tested.

In the final step, the results of a series of hierarchical regression analyses supported the hypothesis that attachment and shame explained a significant amount of variance in the self-reported outcome variables. Attachment style alone and the combined model were not supported in relation to the parent-reported outcome variables. In all regression models, attachment and shame accounted for overlapping and independent variance in the socio-emotional functioning measures. This finding is consistent with the theory that attachment is partially associated with shame.

While some findings are supportive of the hypothesis that attachment and shame will be significantly associated with measures of psychological functioning, as stand alone variables and in combination, there were also several complicating factors.

First, the model was only supported for child-reported measures. The significant relationship between the self-reported outcome measures and self-reported shame measures might be attributable to shared method variance. However, shared method variance is unlikely to have accounted for the significant relationships between the self-reported outcome measures and attachment, which was coded by the researchers. Second, the very high correlation between the shame measures and all socio-emotional measures does question the relationship between these

variables. For example, do the shame measures pick up on the same psychological phenomena as the socio-emotional measures as has been previously been questioned (Del Rosario & White, 2006). Similarly, can shame be conceptualized as a contributory variable or does it belong as an outcome variable. Third, while it was anticipated that attachment and shame might account for overlapping variance in socio-emotional functioning, shame was not expected to be so strongly, and independently, associated with the outcome measures. As such, this may suggest that shame is more likely to act independently to attachment which is at odds with many attachment and shame researchers who believe the two constructs to be inextricably linked (Schore, 1998). Shame acting independently from attachment in relation to socio-emotional functioning may suggest that shame is more strongly influenced by socialising experiences outside of the attachment relationship as hypothesized by other theorists (e.g., Leeming & Boyle, 2004). Finally, without longitudinal data, it is not possible to test causal relationships between attachment, shame and the outcome variables. It may, indeed, be the case that shame is highly associated with psychological function because individuals feel ashamed as a result of their socio-emotional functioning rather than their disfigured appearance.

The literature on variables contributing to psychological functioning in young people with congenital disfigurement is still developing. Existing studies have found some social factors such as peer harassment and social support to be weakly significant contributors (e.g., Feragen & Borge, 2010; Feragen et al., 2010). The ARC study (Clarke et al., 2013) investigating psychological factors contributing to adjustment in disfigured adults referenced attachment and shame in their model but did not directly measure these, categorizing attachment as a factor that cannot be changed therapeutically. However, given the findings of the current study alongside studies on adults with dermatological conditions that show an increased risk of attachment

insecurity (e.g., Rabung et al., 2004; Picardi et al., 2003b; Picardi et al. 2005), attachment style may be worth further investigations. Similarly, there is now a rapidly growing evidence-base supporting the efficacy of attachment-based psychological interventions (e.g., Bakermans-Kranenburg, van Ijzendoorn & Juffer, 2003; Fonagy & Bateman, 2006) which increases the relevance of attachment research in this population. While the focus of research with disfigured adults has focused on cognitive processes that contribute to psychopathology due to the widespread use of cognitive interventions in adult psychological therapies (e.g., Clarke et al., 2013) there is a great deal of evidence to support a range of different interventions in children and families such as behavioural, parent-focused and relational interventions (e.g., Bakermans-Kranenberg, van IJzendoorn & Joffe, 2003; Bernstein, Layne, Egan & Tennison, 2005; Shirk & Karver, 2003; McCart, Priester, Davies & Azen, 2006). As such, explanatory models seeking to identify contributing factors that are amenable to therapeutic change in young people should investigate factors beyond cognition.

5.2.5 Post hoc analysis

A follow-up study was conducted in order to test out recently reported findings that disfigurement visibility significantly influenced psychological functioning in adults with disfigurement (Clarke et al., 2013).

As visibility of disfigurement was not a planned variable, no specific measure of visibility was administered to the participants or their parents. As such, a proxy measure of disfigurement visibility was constructed using information already gained about disfigurement location. Participants were then grouped according to whether their disfigurement was routinely visible to others or not and group means or frequencies on all psychological measures administered as part of the main study

were compared. The results showed that there were no significant differences in psychological functioning according to disfigurement visibility.

These findings are in stark contrast to those reported by Clarke et al. (2013) that were based on an adult sample. However, they were consistent with the findings of a number of studies that investigated children with disfiguring conditions (e.g., Feragen & Borge, 2010) that reported no significant influence of disfigurement visibility. There were a few key differences in the studies that might account for these contradictory findings. The most obvious difference is in the samples of adults versus children. It may be that the impact of having a very visible disfigurement only becomes important when the individual gets older. On the other hand, it may be that, in childhood, it is more difficult to keep one's body invisible. That is, children and young people are more likely to be seen undressed by their parents and other carers, for example. As such, the distinction between visibility and non-visibility of a disfigurement may be very unclear. On the other hand, for adults who have greater control over who they choose to show their bodies to and who have more independence to keep parts of their body non-visible, such as by wearing long sleeves and trousers and not going swimming, the visibility of their disfigurement may take on a different significance. Furthermore, the studies based on children used disfigurement information that was reported by a parent or guardian while the ARC adult study used participant self-report to rate disfigurement. As such, shared method variance may have played a part in the ARC study findings. For example, participants who were functioning less well psychologically may have perceived their disfigurements to be more visible to others via information processing biases that might also underlie their psychological difficulties such as hypervigilance to threat or learned helplessness. In contrast, disfigurement visibility in the child studies was determined by the researcher or, in the case of the current study, by a combination

of the researcher recoding information provided by the parent/guardian of the participant.

5.3 Conclusions

Overall, the study found interesting results. There was little support for the hypothesis that congenitally disfigured young people would demonstrate poorer socio-emotional functioning than their non-disfigured peers. However, the high level of heterogeneity in socio-emotional functioning reported in previous studies found some support in the current study with results indicating better than normal functioning on some measures and worse than normal functioning on others, with the majority of findings indicating no difference in socio-emotional functioning in the disfigured group. As predicted, the disfigured group was significantly more likely to demonstrate insecure attachment style. The findings on shame were contrary to expectations in that the non-disfigured comparison group reported significantly higher levels of shame than the disfigured group. However, the non-disfigured group also reported significantly poorer psychological functioning generally. This may have been an indication that the comparison group was unusually psychologically troubled meaning that they may not have constituted a representative sample of the general population. Analysis of the relationship between attachment and shame showed a weakly significant relationship between attachment insecurity and higher shame. Internal and external shame were so highly correlated that only external shame was included in the final test of the proposed explanatory model. The final set of regression analyses used to test the full, proposed explanatory model of socio-emotional functioning in young people with congenital dermatological disfigurement found that attachment and shame together accounted for a significant amount of variance in the self-reported measures, as expected, but not in the parent-reported measures. The results also indicated that, while attachment and shame accounted for a small amount of

overlapping variance, there was a larger portion of variance in shame and socio-emotional functioning that was independent of attachment. The post hoc study examining the impact of disfigurement visibility on psychological functioning found no support for the hypothesis that young people with visible disfigurement would show poorer psychological functioning than those with non-visible disfigurement.

A key finding that was not a part of the original main research aims was that co-morbid medical variables, were more consistently found to influence socio-emotional functioning. This was more evident in the parent-reported measures, nearly all of which were significantly worse in the presence of the named medical factor and which all showed larger effects than the self-reported measures. On the self-reported measures, Peer Problems was most likely to be significantly worse in the groups with co-morbidities. Pain was the variable that most consistently distinguished socio-emotional functioning measures.

These findings are very important in that they suggest that, in young people with dermatological conditions, any attempt to understand psychological functioning in this population must take into consideration the complex, multi-factorial nature of their medical condition.

In relation to the original, proposed explanatory model of socio-emotional functioning in young people with congenital dermatological disfigurement, within the limitations of a cross-sectional design, there was significant statistical support for the main body of the model: the associations between congenital disfigurement, attachment, shame and socio-emotional functioning, the only change being the amalgamation of internal and external shame into a single shame variable due to the high correlation between them and the qualification of socio-emotional functioning as self-reported socio-emotional functioning since the model was not

supported in relation to parent-reported measures. The results also indicated that shame and socio-emotional functioning appeared to be influenced by factors not tested in the current study. This might indicate that other factors such as social rejection might play a bigger part in the explanatory model than originally thought. The results of the within-group analyses also highlighted the possible influence of other illness variables on socio-emotional functioning in this population, which might mean that disfigurement might act in concert with other illness variables to influence social and emotional functioning. A revised explanatory model of socio-emotional functioning in congenital dermatological disfigurement including new factors that have emerged as potentially interesting, but whose relationships with the original variables is still unclear, is shown in Figure 5.2.

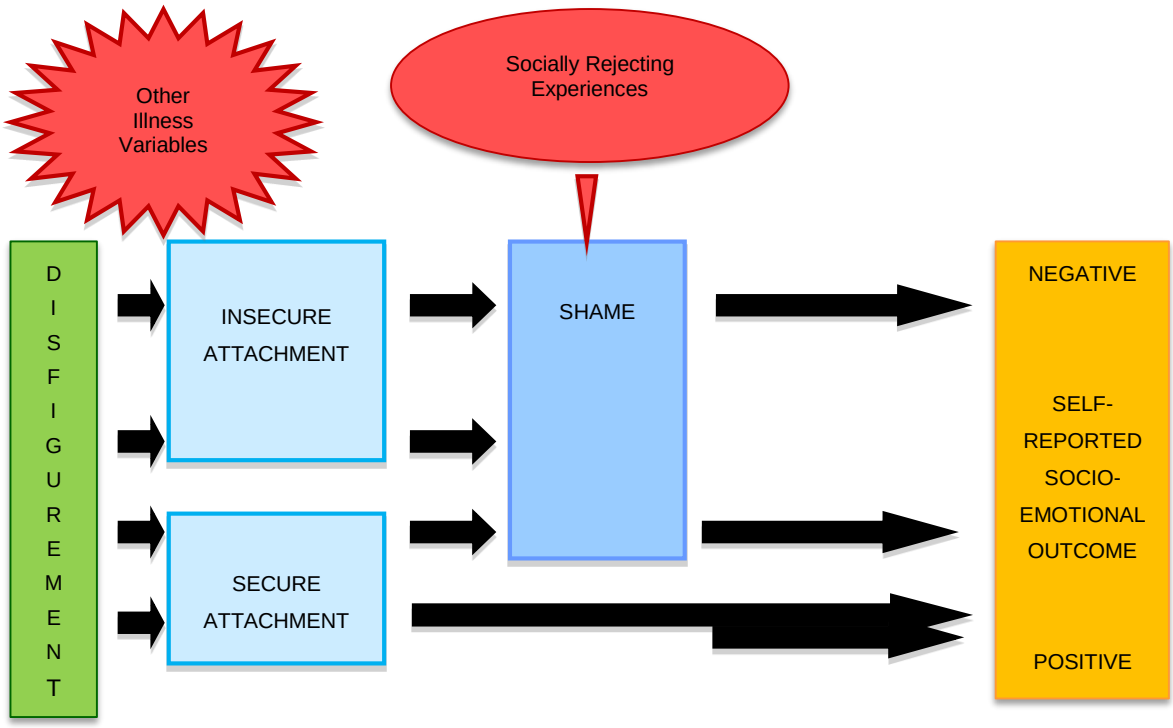

Figure 5.2. Revised Explanatory Model of Socio-Emotional Functioning in Young People with Congenital Disfigurement.

5.4 Discussion Points

A number of conceptual and methodological dilemmas emerged from the execution of the current study. Some of these are discussed below.

1) Does congenital dermatological disfigurement affect socio-emotional functioning in children and adolescents?

The results of this study did not consistently support the hypothesis that children and adolescents with congenital dermatological disfigurement were significantly more likely to experience socio-emotional difficulties than the normal population. This was surprising as the theoretical literature on the impact of unattractive or disfigured appearance on observer reactions is fairly unequivocal. On an empirical level, however, the results of previous studies have been mixed and the findings of the current study are consistent with this mixed picture. There are a number of hypotheses for the unequivocal psychological picture for this population

Congenital dermatological disfigurement and positive psychological development

There are several possible reasons why most disfigured children and adolescents do not experience significantly worse psychosocial functioning than their non-disfigured peers. First, developmental theory suggests that most children of the age range sampled for the current study may not be capable of thinking about the future implications of their disfigurement; a cognitive ability that Piaget named Formal Operational thinking (Piaget, 1976). Piaget believed that this stage of cognitive developmental occurred in middle to late adolescence, although later studies demonstrated a wide variance in when people developed this ability with many not acquiring with this skill until adulthood (Miller, Drotar, & Kodish, 2004; Mulvey & Peeples, 1996). Being unaware of the likelihood that their disfigurement will impinge on most domains of their lives as they get older such as in getting a job and finding a romantic partner may be temporarily protective. Furthermore, while socially rejecting experiences in their childhood may be unpleasant and upsetting, the attributions that disfigured children make about these experiences of rejection may be very similar to those of their non-disfigured peers and not focused on the internal, global and stable reasons for the rejection; their disfigured appearance. Alternately, experiencing more social rejection may be protective in that disfigured children may have developed better means of coping with the rejection such as through more sophisticated cognitive attributions, better skills for managing difficult social situations and have a more sensitized system to support them; teachers and parents being vigilant and responsive to teasing and bullying for example. This last hypothesis has been proposed by earlier study authors who found that psychological functioning in the disfigured group was significantly better than in the non-disfigured comparison group (e.g., Feragen et al., 2010; Andersson et al., 2012).

The differential impact of congenital and acquired disfigurement is still unclear. In context of the current findings, one could argue that children who have always been disfigured may not be able understand how different, arguably easier, their life might be without the disfigurement. The same may be true for their parents and wider system. As such, the disfigurement may have less impact than expected, on a day-to-day basis, on how the people who matter the most interact with these children.

Disfigurement and Unattractiveness

Griffin & Langlois (2006) demonstrated that it wasn't just that "beauty is good" (Dion et al., 1972) but that, also, "ugly is bad". However, the process by which "ugliness" impacts on individuals and functions in the social context may be more complex than the Griffin study indicated. The implicit hypothesis underlying the belief that disfigured individuals are more likely to experience psychological difficulties is that unattractive appearance is presumed to be more likely to elicit negative implicit or explicit reactions from others which would result in signs of psychosocial dysfunction such as in low self-esteem or poor social relationships and ultimately anxiety and depression. However, the experimental studies that have assessed the impact of attractive or unattractive appearance, such as that of Griffin & Langlois (2006), may be flawed in that they have tended to use forced choice response modalities, which may limit the range and complexity of observer response to the appearance of an individual. Furthermore, they have tended not to evaluate the effect of getting to know the unattractive person (e.g., Harper, 1995). It may be that, when faced with a forced choice and no other information about the stimulus individual than their appearance, the observer will rate a more attractive person more positively or select the attract person over the unattractive one. However, some studies (e.g., Lerner, Delaney, Hess, Jovanovic, & Von Eye, 1990) have used responses from observers who know the indexed disfigured person such as the child's class teacher and consistent patterns of preference have been reported.

It may be incorrect to use the findings of studies on unattractiveness to drive our understanding of the impact of disfigurement. Tobiasen & Hiebert (1993) found, in a small study, that general attractiveness, also referred to as background attractiveness, was a separate construct to disfigurement severity in a young CLP population. Indeed, on reviewing the appearance of the participants in the current study, captured on the video recordings of the CAI, there were several participants who, despite having a facial birthmark, large naevus or excoriated skin appearance due to severe eczema, were otherwise quite attractive in appearance on other dimensions of attractiveness. This is consistent with the body of research that indicates that overall physical attractiveness is determined, not just by facial appearance or skin appearance, but by a number of physical indices including body mass index (Tovée et al., 1999), body shape (e.g., Fan et al, 2005) symmetry of features (Fink et al, 2006) and even type of voice (Hughes, Dispenza & Gallup, 2004). It is not clear what impact fashionable clothes or hairstyle might have on perceptions of attractiveness also. As such, it may be possible to be disfigured at the same time as being attractive. Furthermore, a disfigurement that is associated with a medical condition, although it might, on a prima facie level, result in the individual being viewed as unattractive, may elicit a different meaning for the unattractiveness than otherwise unexplained unattractiveness. Socio-anthropological studies have demonstrated that unattractive appearance is strongly associated with negative attributions such as wickedness or lack of intelligence (e.g., Langlois et al., 2000b). However, unattractiveness that is clearly attributable to a medical condition, over which the individual is seen to have no or little control, such as congenital birthmarks, may not carry the same negative personal attributions as general unattractiveness. As such, it may be an inaccurate and oversimplified assumption that disfigurement is equivalent to “ugly” as is assumed in much of the empirical literature.

Is all disfigurement equal?

The term "disfigurement" encompasses a broad range of manifestations across domains such as location, size, appearance, permanence and visibility (to others) of the disfigurement. Furthermore, the cause of the disfigurement, in the case of the current study congenital dermatological disease, can be varied and also cause other symptoms such as pain and mobility problems. It is, as yet, not clear what impact this "biological" heterogeneity had on the results of the current study nor in the existing literature. The post hoc study that divided the sample into a more disfigured and less disfigured group showed some significant differences with the more severely affected group being significantly worse off on parent-rated overall problems, emotional difficulties and peer problems. Overall, there was a consistent trend for all outcomes, except for attachment, to be worse in the more severely affected group. At the same time, significance of difference was not reached for most measures indicating some support for the possibility that the more noticeably disfigured individuals were more likely to have difficulties than the less affected group. However, these results were complicated, not just by the lack of significant differences, but also because there was clearly covariance between the disfigurement-based groupings and other medical factors such as pain, mobility problems and number of hospital visits. Furthermore, participants with eczema were significantly more likely to fall into the more severe category. These co-varying variables call into question whether the differences in outcome measures across the two disfigurement severity groups are, in fact, attributable to difference in disfigurement rather than differences in other, co-morbid, factors.

2) Child versus Parent Report.

While it was expected that there would be differences between child and parent-reported outcomes, hence the importance of collecting both perspectives, the

patterns of outcomes and relationships between outcome measures was interesting. The parent scores were more likely to indicate worse functioning than population norms whereas the child ratings were more likely to be the same as or better than the population norm. As such, overall, the parents seemed to perceive greater difficulties in their child's socio-emotional functioning than the child themselves. This finding is consistent across many different paediatric psychology populations such as children with cancer (e.g., Phipps & Srivastava, 1997) and children with Epidermolysis Bullosa (Soon et al., 2006). Furthermore, the parent-rated measures of socio-emotional functioning related differently to other psychological measures in the statistical analyses. As such, using either parent-rated or child-rated measures can provide a very different psychological picture. The, as yet, unanswered question is which report is more relevant?

Some authors have hypothesized about various processes that might account for the differential reports. For example, parental anxiety about their child's medical condition or residual guilt for causally contributing to the child's illness have been suggested as reasons why parents may report higher levels of concern about their child's psychosocial functioning than their child. Alternatively, the child's desire to appear as normal as possible, to not burden their parents and professional carers further, has been suggested as a reason why the child respondents may have under-reported their own psychosocial problems.

The current study may have provided some further clues to help to understand this phenomenon better. This is due to the inclusion of measures that are not purely based on subjective parental or child reports. These were the CAI and disfigurement severity rating scale, which were rated by at least one, or more, researchers. Some of the descriptive indices of the disfiguring condition, while completed by the parent and therefore somewhat affected by their perceptions and response styles, were

also based in externally verifiable information such as name of disease and number of hospital visits per year as well as the more subjective ratings of symptomatology such as pain and mobility problems.

A pattern emerged in the associations between child reported, parent reported and "other" reported outcome measures. In relation to the independently rated attachment measure, the child reported measures were more likely to be associated with attachment security, than parent measures. Since the existing evidence-base suggests an association between attachment security and psychosocial indices, these results may be interpreted as suggesting that the child reported outcomes are more "psychologically coherent" than the parent reported outcomes. Furthermore, the parent reported outcomes were more strongly associated with the other disease-related factors. These findings might be interpreted to indicate that parental report of socio-emotional functioning was, to some extent, associated with indices of disease severity. That is, parents may be influenced by the common assumption that the more severe their child's medical disease, the more problematic their socio-emotional functioning. Alternatively, more severe medical disease variables may contribute to higher levels of stress or other negative emotions in the parent that might contribute to reports of poorer child socio-emotional functioning also.

Overall, in the context of research about psychosocial functioning in the paediatric population, it is still unclear how to measure psychological function of the child participant in the most accurate way possible. While multiple informant studies have been recommended by several researchers in the field (e.g., De Los Reyes and Kazdin (2005) it is also not clear how to incorporate these measures in a valid and reliable fashion that captures the actual functioning of the participant most accurately.

3) Attachment and Disfigurement

These findings provide a new perspective on disfigurement and attachment. First, there are now a small number of studies that indicate that people with dermatological conditions are at an increased risk of attachment insecurity. Unfortunately, the Rabung and Picardi studies did not suggest a causal pathway by which the dermatological condition was linked to attachment insecurity. Only the current study ventured a possible explanatory pathway; via the disfiguring effect of dermatological conditions. However, as a cross-sectional study, the question of how having a dermatological condition is related to attachment insecurity is still unanswered.

The most commonly cited studies of attachment and disfigurement are those conducted by the Speltz group (Speltz et al., 1997; 2000) on infants with CLP as well as the study conducted by Murray and colleagues (2008) all of which found no association between attachment security and disfigurement. The findings of the current study may have differed from these studies for a number of reasons. First, the difference in attachment security may be a function of the type of disfiguring condition affecting each sample. As such, it may be possible that congenital dermatological disfigurement is associated with attachment insecurity while disfigurement relating to CLP is not. One reason for this differential relationship might be that each disfiguring condition has very different disease courses and treatments. While CLP is associated with significant functional difficulties such as eating and vocalizing, most of these functional problems are resolved when the cleft is repaired at 12-18 months of age. After the repair, there are few functional difficulties and few treatments required for most patients. In the case of the dermatological conditions affecting the current sample, the disease course has been prolonged and most conditions require daily management and recurring invasive medical procedures. Many include functional difficulties and other physical

symptoms and the disfigurement is unresolved. However, in terms of attachment formation, the majority of the current sample was born with their disfiguring condition in the same way as the CLP population. Therefore, attachment insecurity may be associated with disease or disfigurement course.

Second, the different attachment findings may be a function of the age of the sample. The CLP studies focused just on children in the first few years of life while the current study focused on school-aged children and teenagers. The early studies hypothesized that congenital disfigurement might act as a barrier to secure attachment formation between infant and attachment figure(s) (e.g., Wasserman & Allen, 1985). However, it may be possible that increased attachment insecurity occurs over time rather than at the point of initial primary attachment formation in infancy.

Attachment research suggests that this is plausible. There is evidence to suggest that attachment security is unstable in the face of significant life stress (Weinfield et al., 2000). As such, infants with disfiguring dermatological conditions may start life with a normal chance of forming a secure attachment relationship with their primary attachment figures, much like infants with CLP but, over time, in the face of chronic or severe stresses relating to their condition their attachment style changes. The life stress may be in the form of the impact of disfigurement on social relationships, as proposed by this study, or it may be related to the other symptoms experienced by young people with dermatological conditions such as mobility problems which were also found to significantly distinguish attachment categories in the current study. Anecdotal evidence based on clinical practice would suggest that the stresses of managing complex dermatological conditions; the time-consuming, complicated and painful treatments, the limitations placed on family life, the increased demands on parents and the impact of chronic stress on the young person's behaviour and

emotional state can lead to breakdowns in the relationship between the child and the adult primarily responsible for their daily medical care, usually their mother. Based on clinical anecdotal evidence, mothers of chronically ill children often report feeling distraught that they are the person who inflicts pain and distress on their child, in the process of administering treatments, rather than the person who protects their child from pain and distress. This unfortunate result of chronic illness may also affect attachment security in the young person with the dermatological condition.

Differential impact of disease course and attachment instability over time support the importance of studies using concurrent attachment. Longitudinal data measuring attachment in infancy and periodically throughout childhood and adolescence would also help to determine at what point in the developmental lifespan the risk to attachment security occurs in this population.

4) The assumption of social rejection and the reaction to it.

The current study set out to investigate shame as potential contributor to psychological functioning. Shame was considered an important variable to investigate because it is defined as the emotional response to social rejection (e.g., Gilbert & Andrews, 1998). Social rejection has been long considered to be a key stressor for young disfigured people because of the important role of appearance in social relationships.

An “objective” measure of social rejection was not used in the current study because the existing research on social rejection has indicated that the experience of social rejection, even an agreement on when social rejection has occurred, is highly subjective and, therefore, very difficult to reliably define and quantify (e.g., Olweus, 1994). The current study aimed to take into consideration this transactional process

by measuring the subjective emotional outcome of social rejection: shame. However, shame was not found to be elevated in the disfigured population as expected. This outcome may have suggested that 1) disfigured children were not experiencing increased social rejection as assumed 2) their responses to the increased social rejection were different to non-disfigured populations 3) the measure was not able to pick up on shame responses in this study's population.

The first hypothesis, that young people with a disfigured appearance might not be experiencing increased social rejection, is plausible in that, as already discussed, "unattractive" appearance that is obviously illness-related may not elicit the same negative attributions as other forms of unattractiveness such as a high BMI, which has typically been associated with undesirable personal traits such as laziness and greed (Puhl & Heuer, 2009). On the contrary, disfigurement that is clearly associated with medical illness may attract pity or sympathy and consequently overly inclusive or solicitous behaviour (e.g., Maris et al., 2000). Similarly, overtly rejecting a "sick" peer might be seen as less acceptable than rejecting another low status child such as an overweight child or a socially awkward child. As such, a child with a known or identifiable medical condition may not receive as much overt social rejection as other children considered to be of low social value. However, anecdotally, this first hypothesis seems unlikely given that social problems relating to disfigurement are the most common referral to paediatric dermatology psychosocial services (Soon, Creese, Marks, & Jeffries, in prep).

The second hypothesis for not finding higher levels of shame in the disfigured sample is that this population has an idiosyncratic reaction to social rejection such that their responses are not typical of the general population and do not result in elevated shame. Again, there is some plausibility in this rationale. It may be that the parents of disfigured children are primed to respond actively to socially rejecting

experiences because their child is seen to be vulnerable. This is in keeping with the conclusions of some studies that suggest that the parents of disfigured and/or sick children are more emotionally attentive to their child than parents with healthy children (Maris, 2000). This is also supported by the finding that disfigurement-related concerns are a common referral problem (Soon, Creese & Jeffries, in press). Parents and carers may be more likely to actively and pre-emptively address the issue of social rejection by peers because they are aware of their child's difference. This might take the form of socialising their child into how to manage these situations themselves, by liaising with the school and working to increase social inclusion and to ensure that teachers respond robustly to any signs of social rejection or asking for referrals to mental health or social care professionals for help for their child in managing these experiences. Parents, and others in a caring role, may also be more likely to actively resolve episodes of social rejection because of their concern about the impact on their already vulnerable child. Ultimately, a more active response against social rejection may implicitly signal to the child their social importance, at least to those people closest to them. Directly and indirectly, a more robust approach towards addressing social rejection may help the disfigured child to externalize responsibility for socially rejecting experiences or to defend themselves in the face of overt social rejection. It may be interesting to compare medically-related disfigurement against other forms of "unattractiveness" such as obesity or non-medical unattractiveness in relation to response to social rejection.

It may be the case that children and young people may not be as aware of social rejection as their adult counterparts who are more likely to report higher levels of psychosocial dysfunction (e.g., Picardi et al., 2003b). Children and adolescents might also not apply the same meaning or attributions to socially rejecting behaviour as disfigured adults because of developmental differences. Finally, given that the recent literature indicates that children and young people in the non-disfigured

population report high rates of social rejection, such as in the form or bullying (e.g., Lovegrove & Rumsey, 2005) it may be that disfigured children and young people perceive their own socially rejecting experiences as being part of a universal social experience and, therefore, are less likely to internalize or personalize the negative messages. As such, the disfigured child's cognitive, behavioural and emotional response to social rejection may be different to other children in a number of different ways. However, there is currently no empirical evidence to support this idea.

The final hypothesis, that the shame measures used in this study were not able to access the shame experience in this population in a valid and reliable way is also plausible. The measures used have been well-validated on adult populations with, typically, fairly high levels of psychological vulnerability. However, they have not been validated with child or adolescent populations. While the wording of some of the questionnaire items was changed, as per the recommendations of the research ethics committee that reviewed this study, to make each shame questionnaire more understandable to children and adolescents on a verbal level, it is not clear if the internal and relational phenomena described in the questionnaire items were at all recognizable to young people or if, in fact, valid to the social functioning and cognitive processing of children and adolescents. However, the high correlations between shame scores and scores on other better-validated child-reported measures suggested that the shame questionnaires were accessing something akin to psychological and social problems although it is unclear if the questionnaires were accessing what the questionnaire and this study's authors intended. As such, it would be helpful for future research to more specifically describe the shame phenomenon in children and adolescents. Shame is a construct that manifests in different domains and in different ways. It may be that there are more accurate,

reliable and valid ways of accessing the experience of shame in younger populations.

A further measurement issue is the impact of one shame-related behaviour, which is concealment. It may be possible that children who experience exceptionally high levels of shame or who experience shame about feeling shame are more likely to conceal, from their own awareness and/or from others, the true nature and level of their shame experience.

While lower shame may be interpreted as a positive outcome: that adults should, perhaps, be less concerned about the impact of negative social experiences on this group of young people, an alternative perspective is that shame, being a socially facilitating phenomenon, is worryingly absent from this population. An inability to experience shame in a normal way may reduce a child's likelihood to develop inclusive social behaviours and to avoid socially inappropriate behaviours thereby contributing to further social isolation.

5.5 Limitations.

There were some methodological limitations that should be considered when interpreting the results of this study and in considering future research and clinical practice.

5.5.1 Sampling.

There are a number of factors that call into question the representative nature of the sample on which this study is based.

First, participants were sampled from specialist clinics in a well-known, highly specialized, paediatric hospital in the UK. Drawing participants from specialist

services is very common in studies on disfigurement in childhood, presumably because of the ease of access to large numbers of potential participants with relatively rare disfiguring conditions. Few studies have sampled a similar population from community sources. As such, it is unclear if patients attending specialist services, that are often highly resourced and able to provide a high level of medical and psychosocial support to the patient and their family, are psychologically similar to patients and families with community-based, generalist support.

Second, the final sample constituted a relatively small proportion of the number of patients approached. Five hundred and sixty six families were approached about the study but only 122 were eventually included. Of those who did not participate, 79 elected not to participate because they did not feel it was relevant to their child or because they were concerned about the negative impact of discussing these issues with their child. Within this group there may have been some children and families who were the most negatively affected by their disfigurement, so much so that they felt unable to discuss their experiences openly. While it is impossible to ascertain systematic psychological differences between participants and non-participants, it is unclear if the final sample was appropriately representative of children and adolescents with congenital dermatological disfigurement.

The third sampling factor, which may have affected the results, was sample size and missing data. The original power calculation estimated a sample of 86 for adequate statistical power. While 122 participants were included in the final sample, because of high amounts of missing data only 79 data sets were included in the multiple regression analyses on which the power calculation was based. As such, the most complex analyses may have been slightly underpowered.

Fourth, another sampling issue for the current study was that disfigurement severity was not controlled for in the main study. The subsequent post-hoc study looking more closely at the role of disfigurement severity found some significant differences between the most severely disfigured participants and the remainder of the group. Even when between group differences did not reach significance, there was a consistent trend for the severely disfigured group to score more poorly than the general group. While disfigurement severity co-varied with other illness variables, if the study had applied a higher threshold for inclusion based on disfigurement severity, the results may have been more robust compared to those currently reported.

5.5.2 Design.

The major limitation of the current study was it's cross-sectional design. A test of a causal model, inherently requires a longitudinal design in order to fully test the causal directions of the relationships between variables. However, because conducting a large-scale longitudinal study is a significant research undertaking, it made practical sense to conduct a cross-sectional study first to determine if there was evidence of the proposed relationships between variables. As such, the current study was a preliminary step in testing the proposed explanatory model that will, hopefully, be tested more robustly using a longitudinal design.

5.6 Implications for Future Research

The results of this study posed nearly as many new questions as it answered old ones. A number of points were highlighted that should be considered or addressed in future research.

Other Medical Factors

Conducting research on a sample defined by the term “congenital disfigurement” may not be sufficiently specific to properly understand the psychological developmental pathways in young people with congenital disfigurement. The current study indicated some heterogeneity within the disfigured sample relating to the other features of the medical condition underlying the disfigurement. Other disease variables, such as pain, appeared to be more powerful in differentiating some indices of socio-emotional functioning in the participants than disfigurement. The complex symptomatology relating to different congenitally disfiguring conditions may result in difficulty in generalizing findings across disfigurement samples and may call for a higher level of control over co-morbid symptoms when selecting and describing samples. It will be important to try to titrate out the relative effects of disfigurement and comorbidities such as mobility problems, pain, disease course on psychological adaptation.

Social Rejection

While the study findings supported the hypothesis that attachment and shame would be associated, the results indicated a weakly significant relationship. Furthermore, the regression analyses indicated that a greater proportion of variance in shame and socio-emotional functioning was accounted for by factors other than attachment. A number of other factors have been theorised to influence shame. These include stigma, social rejection and devaluation outside of the primary attachment relationship and parenting and family styles. All of these factors have face validity as potential contributing factors to shame and socio-emotional difficulties in young people with disfigurement and some have already been studied in relation to disfigurement. The results of the current study suggest that further research in this area might be useful in further developing the causal pathway between congenital disfigurement and socio-emotional functioning.

It may also be helpful to examine differences in people's reactions to unattractiveness and to disfigurement. Research has determined that a number of different factors contribute to the evaluation of an individual as attractive or unattractive. As such, disfigurement and attractiveness may not be mutually exclusive. Furthermore, unattractiveness caused by a known medical condition may elicit different attributions and, therefore, social actions, compared to unattractiveness that is not easily or "externally" attributable. As such, the meaning and impact of disfigurement related to a congenital medical condition could usefully be explored to determine if it elicits a different set of reactions to other causes of unattractiveness. This issue of social response to disfigured people underpins much of the research conducted with young people with congenital disfigurement. However, this phenomenon of social stress, relating to having a disfigured appearance, is still not unequivocally proven.

The mixed findings in the literature, including this study, regarding social functioning in disfigured young people and the unexpected findings relating to shame in the current study indicate that it might help to test specific stages of the underlying hypothesis – that disfigurement results in social rejection which results in psychological stress which results in measurable socio-emotional difficulties. There may be fundamental flaws in these causal assumptions that have contributed to the mixed, equivocal findings on social functioning in this group of young people.

Other Illness Variables

An unexpected finding was the impact of medical variables on socio-emotional functioning. Indeed, most of the medical variables measured demonstrated a more consistent and powerful distinction in socio-emotional functioning than the key variable of disfigurement. As such, further research on psychological functioning in

young people with disfiguring dermatological conditions should, at least, incorporate medical variables into the model, if not investigate them directly.

Parent versus Child Report

It may be helpful to explore the relationships between parent and child-reported outcomes. Ongoing contradictory findings because of reporting discrepancies will only obstruct further investigation of child populations, potentially silencing the perspectives of children and their opinions about their own bodies, feelings and lives as well as slowing down the development of effective clinical practice for child and adolescent populations.

5.7 Implications for Clinical Practice.

The primary purpose of this study was to contribute to the development of better clinical interventions for young people who are experiencing psychological difficulties relating to their congenital dermatological disfigurement. While the research is in its early stages, some points emerged from the results that may be useful to hold in mind when providing mental health services for this population.

One clear pattern to emerge from the current study was a high level of heterogeneity in the reports of socio-emotional functioning in children and adolescents with disfiguring dermatological conditions. As such, clinicians should be very careful with how they assess for psychological difficulties with this group. First, psychopathology should definitely not be assumed in this population. Second, clinical assessments should take into consideration differences in parent and child reported difficulties. Third, even when socially rejecting experiences are confirmed, it may be important to check, carefully, the child's own interpretation of that experience. Furthermore, despite disfigurement being the most common clinical referral problem in children with severe dermatological conditions, more attention

should perhaps be paid to some of the other key medical variables in dermatological disease when considering psychological vulnerability in patients.

In terms of the main findings of this study, psychological domains most likely to be problematic in young people with congenital disfigurement appear to be internalising problems and peer relationship difficulties. The current study also found some evidence for elevation in total scores of psychopathology. This fits well with clinical anecdotal evidence of common difficulties faced by this group of young people. As such, clinical interventions that are specifically focused on these areas of concern might improve the efficacy of clinical psychology interventions. There is also evidence of increased risk of attachment insecurity in this population and that attachment security may be linked to the socio-emotional difficulties that the young person is presenting with. As such, incorporating attachment processes into the clinical assessment and intervention may be useful in shifting the presenting problem. Furthermore, because attachment insecurity can have a negative impact on engagement in therapy, the clinician might usefully be mindful of this in order to ensure that the young person and their parents are facilitated into maintaining a positive working relationship with the clinician. Finally, there appear to be several factors that may be significantly associated with socio-emotional functioning in this group of young people, physical/medical factors, attachment and shame being just some of them. As such, clinicians should hold a multi-factorial explanatory framework in mind as they seek to understand the impact of congenital dermatological disfigurement on the young person before them.

5.8 Final Summation

Although the way in which young people are taught to value their own bodies and their appearance and the impact of heavily skewed media images of ideal body types and of beauty are important socio-political topics currently and have attracted a great deal of research interest, robust empirical investigation of young people with disfigured appearance is still developing. Further, the empirical literature on children and adolescents with congenital dermatological disfigurement is even more limited. This lack of empirical evidence presents a challenge to clinicians in the field where psychological and social difficulties relating to disfigured appearance are a common presenting problem.

The current study aimed to start to address the limitations in the existing literature by describing the socio-emotional needs of the population as well as trying to identify developmental variables that may contribute significantly to socio-emotional functioning in disfigured young people. It was hoped that the findings of this study would provide clues as to how to work clinically with disfigurement-related psychological difficulties.

The study succeeded and failed in fairly equal parts in this endeavor. While the socio-emotional functioning of this population has been consistently shown to be highly variable, the study did provide clues as to why this may be by drawing attention to the medical heterogeneity of this population as well as the heterogeneity between parental and child reports of social and emotional functioning. This study provided new findings in the significant differences in attachment security, in contrast to previous studies and the significant associations between attachment, shame and socio-emotional functioning. This study also highlighted further potential key contributing factors of other illness variables and social rejection.

Future research should examine psychological functioning in this population to a higher level of specificity and explore the specific processes by which attachment, shame and other key variables contribute to individual developmental pathways in order to inform the best possible healthcare provision so that each child is able to achieve their full potential in life regardless of their appearance.

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Appendices

Appendix A

Literature Review: Review Protocol

A.1 Search Protocol

Systematic review of papers on "What is the psychological and social functioning of 8-16 year olds with congenital disfigurement?"

OBJECTIVE

1. To compare the population of 8-16 year olds with congenital disfigurement with the general population or other clinical populations on the same indices of psychosocial functioning.

A.2 METHOD

The "PICO" structure, standing for Participants, Interventions, Comparators and Outcomes, recommended by the Cochrane Collection (2008) and the Centre for Reviews and Dissemination in York (2008) was used as a framework for the systematic review. The PICO for the current review is as follows:

P: Eight to 16 year olds with congenital disfigurement.

I: There are no interventions as this is a review of descriptive studies.

C: The general population in the form of published norms or non-disfigured comparison groups drawn from the general population or non-disfigured clinical populations.

O: Psychological, including social but not intellectual functioning, in 8-16 year olds with a congenital disfigurement.

A.2.1 Criteria for considering studies for this review

A.2.1.1Types of Studies

Papers that compare the data drawn from the indexed population against other population data using quantitative methodologies will be included.

Opinion papers, review papers, qualitative and single case studies will be excluded.

A.2.1.2 Types of Participants

The indexed participant will be boys and girls who are 8 – 16 years of age (including studies where 50% or more of sample is between 8 and16 years of age). They will have been diagnosed with a congenital disfigurement and will have been sourced from clinical or non-clinical settings. Studies that consist solely of populations known to have a high frequency of learning disabilities and studies with a sample consisting of more than 50% of participants with identified learning disabilities will be excluded from the review. People with learning disabilities are known to have significant psychological and social difficulties that directly relate to their learning difficulties. As such, co-morbid learning disability, which is the case in some disfiguring syndromes such as Aperts Syndrome and Neurofibromatosis, may significantly skew the psychological picture of the disfigured population.

Data will be self-reported or reported by parents, teachers or health care professionals who know the indexed participant.

A.2.1.3 Types of Comparators

It is expected that the most common comparators will be the general population in the form of published norms or in the form of comparison groups drawn from the general population.

It is expected that some papers may also compare the indexed population against other clinical populations such as children and young people with acquired disfigurement or non-disfigured clinical populations such as children with chronic illness or mental health disorders. This data will be reported separately.

A.2.1.4 Types of Outcomes

The focus of this review is on studies that describe the psychological and social functioning in the described population. As such, the outcome measures will be any index of psychological or social functioning including indices of mental health disorder such as anxiety and depression, indices of psychological functioning such as attachment, mood, quality of life, self-esteem and indices of social functioning such as social skills, social inclusion and peer relationships.

The outcome data will be provided via questionnaire-based data that has been quantitatively analysed.

A.2.2 Search Methods for Identifying Relevant Studies

Electronic Searches

Relevant studies, published from January 1st, 1990 to April 1st, 2013, will be identified from:

Pubmed

PsycINFO

PsycEXTRA

Web of Science

CINAHL Plus

The search terms used for each database are outlined in the Search Protocol in Appendix X.

A.2.3 Searching other resources

A.2.3.1 References from published studies

A citation and reference search from the papers identified in the core search will be performed to look for any additional, relevant studies that also fit the core search criteria.

A.2.3.2 Unpublished literature

A search of unpublished dissertations has been included in the formal search strategy. Attempts will be made to access relevant unpublished studies via direct contact with the author of these identified unpublished studies.

A.2.3.3 Language

Non-English studies will be included as long as the abstract is in English. Translations of the full paper will be sought as appropriate.

A.2.4 Data Collection and Analysis

A.2.4.1 Study Selection: Phase 1

The researcher will review titles and then abstracts generated by the search and eliminate studies systematically due to duplication, irrelevance or because they do not meet the inclusion criteria. A study attrition flow diagram will be constructed contemporaneously.

A.2.4.2 Study Selection: Phase 2

The researcher will access and read all papers remaining in the review. Further papers may be eliminated due to duplication, irrelevance or for failing to meet inclusion criteria. Information from the papers included in the study will be entered onto the data extraction table and assessed for quality based on the criteria below.

A.2.4.3 Data Extraction

Data will be extracted systematically from each paper using a data extraction proforma (Appendix X), which was adapted from the data extraction checklist described in the Cochrane Handbook (Reeves et al., 2008). Data will be checked and entered. The researcher will contact study authors if further clarification of the study is needed.

A.2.4.4 Meta analysis

It is not anticipated that there will be sufficient studies of an equivalent nature to enter into a meta-analysis.

A.2.4.5 Analysis

The participants, outcome measures and outcomes will be summarized. The methodological quality of the studies will be compared based on the criteria listed in the section titled Quality Assessment.

A.2.4.6 Quality Assessment

The quality of the studies included in the search will be evaluated according to the following indices sourced from the Cochrane Handbook (Reeves et al., 2008) :

1) Adequate Sample Size: This will be determined by identifying the statistical analysis used and calculating the sample size required for adequate power.

2) Representative Sampling Method: This will be determined by evaluating whether there were systematic biases in the way in which participants were sampled.

3) Validity and reliability of outcome measurement: This will be determined by whether or not the study utilised outcome measures with good reliability and validity data.

4) Validity of comparison group: The comparison group will be evaluated in terms of systematic differences to the clinical group as well as sampling biases in order to determine potential sources of error in between group comparisons.

5) Appropriateness of statistical analysis: The statistical analysis used will be reviewed to ensure that it is the most appropriate means of analyzing between

group differences given the nature of the data, the sample size and the question being tested.

6) Reporting biases: Papers will be checked to ensure that the conclusions reported by the authors are appropriately supported by their results.

7) Any other biases.

Appendix B

Literature Review: Search Strategy

Table B.1.
Search Strategy and Results

	Pubmed	**PsycINFO**	**Web of Science**	**CINAHL Plus**	**PsycEXTRA**
Search 1 "disfigurement"	#1: Disfigur*	#1: Disfigurement #2: Disfigurement (MtSH) exp Physical Disfigurement/	(Lemmatization On) #1: Disfigurement #2: Deformity #3: Congenital Abnormality	#1: Disfigurement #2: Deformity	#1: Disfigurement #2: Disfigurement (MtSH) exp Physical Disfigurement/
		#3: Combine #1 OR #2	#4: Combine #1 OR #2 OR #3	#3: Combine #1 OR #2	#3: Combine #1 OR #2
Search 2 "psychological effects"	#2: "psychiatry and psychology category [Mesh]"	#4: Social functioning (MtSH) exp Social Skills/ or exp Social processes/ or exp Social Adjustment/ or exp Social Interaction/ #5: Psychological effects (MtSH) exp Emotional Adjustment/ or exp Stress/ or exp Mental Health/ or exp Psychosocial Factors/ or exp Emotional States/ or exp Well Being/ or exp Psychological Development/	#5: psychological effects #6: social functioning #7: mental health #8: psychological development #9: emotional adjustment	#4 (MH "Adaptation, Psychological+") #5 Mental Health #6 (MH "Social Behavior") OR (MH "Social Adjustment") OR (MH "Social Anxiety Disorders") OR (MH "Social Behavior Disorders")	#4: Social functioning (MtSH) exp Social Skills/ or exp Social processes/ or exp Social Adjustment/ or exp Social Interaction/ #5: Psychological effects (MtSH) exp Emotional Adjustment/ or exp Stress/ or exp Mental Health/ or exp Psychosocial Factors/ or exp Emotional States/ or exp Well Being/ or exp Psychological Development/
		#6: Combine #4 OR #5	#10: Combine #5 OR #6 OR #7 OR #8 OR #9	#7: Combine #4 OR #5 OR #6	#6: Combine #4 OR #5
Combine Searches	#1 AND #2	#3 AND #6	#8 AND #14 AND #22	#3 AND #7 AND #10	#3 AND #6
Results	**150**	**246**	**1989**	**56**	**9**

Appendix C

Literature Review: Data Extraction Form

DATA EXTRACTION FORM **STUDY ID:**

PART ONE: ELIGIBILITY

Type of Study

Qualitative

Methodology of Synthesis

Quantitative

Descriptive/Comparative /Other

Participants

Do the participants in the study have a dermatological condition?

YES UNCLEAR NO

Are at least 50% of the participants between the ages of 8 and 16?

YES UNCLEAR NO

Has the data been elicited from the indexed patient, their parent, a teacher or healthcare professional?

YES UNCLEAR NO

Did the study measure psychological function in the form of mental health disorders, specific areas of psychological function, specific areas of social function

YES UNCLEAR NO

If you have answered NO to any of the questions about participants or outcomes please STOP HERE. If you have answered YES for all questions, please proceed to Part 2.

PART 2: INFORMATION ABOUT THE STUDY

Characteristics of the study

Country where the study was conducted

How were participants sourced? E.g., specialty clinic, general practice, general community

Was the study funded and how?

Characteristics of the participants

Inclusion criteria (please describe)

Exclusion criteria (please describe)

Number of potential participants (i.e. those approached for inclusion)

Number who did participate

Were responders and non-responders compared

Demographic Characteristics

Age range (mean, S.D.) of participants

Gender – number/% female and male

Ethnicity of participants

Socioeconomic status of participants

Disease characteristics

Dermatological Diagnosis

Age of onset

Areas affected

Co-morbidities

Characteristics of Comparison Group (other disfigured, non-disfigured illness, general population, none)

Demographic Characteristics

Age range of participants

Age of participants (mean, S.D.)

Gender – number/%male

Ethnicity of participants

Socioeconomic status of participants

Disease characteristics (if any)

Diagnosis

Age of onset

Areas affected

Co-morbidities

Outcome Measures

What was/were the outcome(s)?

How was outcome assessed (questionnaire, observation, interview etc)

Who completed the outcome measure (patient, parent, other)

How were the outcome data obtained? (face-to-face, telephone interview, postal, other)

Place of outcome assessment (inpatient, outpatient, home)

Results of outcome data

PART THREE: Study Quality

For all studies:

Were hypotheses stated prior to the start of the study?

Were all aspects of the study conducted prospectively?

How were the patients selected? (convenience sample, all patient from data base included etc)

Were measures used for outcome assessment reliable and valid?

Were confounding factors considered? If so, which?

What methods were used to control for any confounding?

Was the method of analysis (qualitative and quantitative) adequately described and appropriate to answer the research questions?

For Comparison Studies:

Was the sample size adequate for statistical power in comparison studies?

Were comparison groups appropriate (size, population, demographics) for the study?

Any further comments about this study?

Is further information required from the authors? YES NO

If yes, give details:

Appendix D

Literature Review:
Additional findings reported by studies included in the literature review

Table D.1
Extra Findings in the Literature Review

Identifiers		Population Studied			Design			
Authors	**Publication Year**	**Age Range**	**Sample Size**	**Type of disfigurement**	**Questionnaires used**	**Reported by**	**Comparison Group**	**Other findings**
Pope & Ward	1997	11- 13 (M=12.7)	24	CFS, CPO, CLP	SPP CBCL	child parent	Published norms	**None**
Shute, McCarthy, Roberts	2007	11-14 (M=12.6)	48	CFS, CLP, CPO	SPP SASA CBCL ,	child Parent	Published norms	**Disfigurement:** indices not associated with outcomes **Age:** Significant inverse association with Harter PA and Hater GSW
Richman & Millard	1997	12	44	CLP, CPO	BPC	Parent	School group	**Disfigurement:** indices not associated with psychological outcomes
Sheerin et al	1995	7-15 (M=11.0)	79	PWS, PE	Harter, RCMAS, CDI CBCL	Child parent	Matched school group, published norms	**Condition:** PE group sig higher than PWS group on internalizing and externalizing problems, overall distress **Gender X Age:** Internalizing disorders and conduct problems significantly worse in older girls
Campis, DeMaso & Twente	1995	6-12 (M=8.1)	77	CLP, CFA, VA	CBCL	parent	Published norms	**Disfigurement:** Maternal adjustment more strongly associated with child outcome than disfigurement indices **Parent:** Maternal adjustment not sig different to norm
Leonard, Dwyer Brust, Abrahams& Sielaff	1991	8-18 (M=12.0)	105	CLP, CLO, CPO	Piers-Harris	Child	Published norms	**Gender X Age:** Adolescent girls significantly lower than younger girls Adolescent boys significantly higher than younger boys
Topolski, Edwards, Patrick	2005	11-18 (M= not specified)	56	CLP, CFA, AD	CDI, YQOL-R	child	1) community 2) ADHD 3) mobility	**None**

							disability	
Andersson et al	2011	9-11 (M=10.6)	92	CHD	Piers-Harris	child	school	**Disfigurement:** Mild disfigurement group significantly lower than severe group on behaviour, intellectual and school status
Feragen et al	2010	16	289	CLP (visible and non-visible)	HSCL-25, Harter	child	Epidemiological sample	**Disfigurement X Gender:** Girls in non-visible group significantly higher appearance satisfaction than Girls in visible CLP and comparison groups **Appearance Perception:** Association between social acceptance and distress mediated by appearance perception
Kapp-Simon et al	1992	10-16 (M=12.3)	45	CLP, CFA	Harter PIC, RBPC	Child parent	Published Norms	**Condition:** No sig differences between CLP and CFA groups
Millard & Richman	2001	8-17 (M=11.7)	65	CLP-U, CLP-B, CPO	RCMAS, RCDS/RADS, CogAT PBS	child parent, teacher	CLP-U, CLP-B, CPO, published norms	**Condition:** CPO sig higher on anxiety and depression than CLP-U, CLP-B. CPO sig higher on learning problems than CLP-U, CLP-B
Padwa et al	1991	6-16 (M=not specified)	30	CFA, CFS	CDI, HFD, TED CBCL	Child Parent, teacher	Symmetrical CFA, Assymetrical CFA, published norms	**Disfigurement:** Symmetrical group sig lower than asymmetrical group on all outcomes **Projective tests**: indicate poor self-concept denial of socialization problems

Appendix E

Comparison Group Data Collection: Method

E.1 Introduction

The current study set out to test two hypotheses relating to shame and congenital dermatological disfigurement in young people.

1) Young people with congenital dermatological disfigurement would report significantly higher levels of shame than the non-disfigured general population.
2) Shame would be significantly associated with socio-emotional functioning in this population.

Because population norms do not currently exist for the measures of internal shame and external shame, the ISS (Cook, 1996) and OAS (Goss et al., 1994b) respectively, data reflecting shame in the non-disfigured general population was required in order to compare shame in the disfigured group with shame in the general population.

In order to create an age-matched comparison group from the general population, primary schools were contacted in order to find a comparison sample of 8 to 11 year olds (year 4, 5 and 6) and secondary schools were contacted to provide a comparison sample for 12 to 16 year olds (year 7, 8, 9 and 10). Both shame questionnaires plus the self-reported SDQ Total Difficulties subscale were administered to pupil participants, during school hours, in collaboration with school staff.

E.2 Method

E.2.1 Sample

Inclusion Criteria

- Boys and girls aged 8-16 years of age.

Exclusion Criteria

- Significant learning disability
- Unable to read or write in the English language to an 8 year old level

E.2.2 Sampling Procedure

- Participants for the comparison group were sampled from three schools in the Greater London area.
- Schools were identified, in the first place, from a list held by the Great Ormond Street Hospital Children's Charity of schools who had expressed an interest in supporting the hospital and the charity. When this list had been exhausted, schools in the local area were identified by checking the Yellow Pages telephone book.
- Schools were telephoned by the researcher and the study was described to the school representative, most commonly the school deputy principal.
- If the school representative expressed an interest in participating in the study, a Schools Information Sheet (Appendix FF) was mailed to them.
- Once the school had confirmed their participation, the procedure for collecting the data was negotiated with the school representative, in order to ensure scientific integrity but also to minimise disruption and inconvenience to the school.

E.2.3 Data Collection Procedure

- All three schools chose an "opt out" consenting process in which parents were sent an Information Sheet about the study which also informed them

that their child would be included in the study unless they contacted the school representative to exclude their child from involvement.

- Parents were given approximately two weeks to contact the school.
- Pupils were asked to participate on the day of data collection and written assent was acquired from each participating pupil.

E.2.3.1 Primary School Data Collection

- A government primary school in North London was the collection site for data for 8-11 year olds. It was agreed with the school representative that the information sheet would be sent by the school to all parents of children in years 4, 5 and 6.
- Because of the young age of the children, the school requested that the questionnaires be completed in groups of approximately 10 pupils that were facilitated by the researcher.
- In turn, participating children left their class in groups of 10 for the separate room in which they completed the questionnaires with the support of the researcher.
- The questionnaires took approximately 30 minutes to complete in this format.
- No concerns or problems were reported by the school, the participants or the researcher coordinating the data collection.

E.2.3.2 Secondary School Data Collection

- Data from two secondary schools were eventually included in the study. The first school, a government comprehensive secondary school in North London agreed to participate

- The school chose to use the same opt-out consenting procedure as the primary school. The school sent the information sheet to the parents of pupils in the appropriate year groups.
- Due to a misunderstanding in the instructions provided by a member of the research team, only one Year 9 form room class (N=33) was included in the study.
- As such, a second North London government comprehensive secondary school was approached in order to collect further data sets from a broader age range of pupils.
- The second secondary school chose to use the same “opt-out” consenting procedure with the parents of the pupils. The school also chose to send the information sheet to the parents themselves.
- Pupils were asked for written assent to participate at the time of data collection.
- One form room class from each year 7, 8 and 9 was randomly chosen to participate in the study.
- Each form room class consisted of approximately 35 pupils from the year group. The form room classes were not streamed in any systematic way and were, therefore, considered to be a random selection of pupils from within each year group.
- Each form room class was given the questionnaires to complete as a single group during a form room period.
- The data collection was overseen by the form room teacher alone.
- The form room teachers involved had been carefully instructed on how to administer the questionnaires and how to minimise bias in the data i.e., to explain the instructions for the questionnaires accurately, for pupils to complete the questionnaires independently, to ensure that all pupils had adequate time

to complete the questionnaires, to provide support where necessary, to ensure completion of the questionnaires and to ensure that anonymity was preserved.

E.2.4 Data Entry

- Raw data was initially entered into a single, separate SPSS datafile.
- Participant datasets were excluded if missing data meant that subscales could not be calculated and if all item scores were the same number (these were assumed to be inaccurate results as all questionnaires administered included reversed items).
- Once the data was considered sufficiently clean, it was added to the experimental group data in the main SPSS datafile for comparison analyses.

E.3 Results

E.3.1 Sample Characteristics

The characteristics of the disfigurement group and the comparison group are shown in Table E.1.

Table E.1
Demographics: Disfigurement and Comparison Group

	Disfigurement	Comparison
Age		
Mean Age	146mth (12yrs-2mth)	133mth (11yrs-0mth)
Age Range	8-0 -16-11	8-0 - 15-11
School Stage	n(%)	n(%)
Primary	50(42.3)	84(71.2)
Secondary	68(57.6)	34(28.8)
Gender	n(%)	n(%)
Female	74 (60.7)	59 (50)
Male	48 (39.3)	59 (50)
Occupational Category	%	%
Higher Professional/Management	29.6	26.2
Lower Professional/Management	24.6	26.2
Technical/Craft	9.8	16.8
Lower Supervisory	9.0	4.7
Semi routine	8.2	8.5
Intermediate	3.3	9.1
Routine	5.0	5.9
Unemployed	5.7	5.1
Unknown	4.8	2.8
Ethnicity	%	%
White British	73.8	45.5
White Other	12.3	18.6
Mixed Race	4.9	4.8
Asian	3.2	18.5
Black	1.6	7.7
Other (inc. Arab)	0	4.9
General Psychological Functioning	X(SD)	X(SD)
SDQ Total Difficulties	10.42(5.9)	17.81(5.4)***

Appendix F

Letter Confirming Ethical Approval

28 APR 2009

NHS
National Research Ethics Service

Institute of Child Health/Great Ormond Street Hospital Research Ethics Committee
The Institute of Child Health
30 Guilford Street
London
WC1N 1EH

Telephone: 020 7905 2620
Facsimile: 020 7905 2201

21 April 2009

Full title of study: **Towards a model of psychological adaptation in children with congenital disfigurement: The relationship with parental mental health, attachment and shame.**

REC reference number: **09/H0713/19**

The Research Ethics Committee reviewed the above application at the meeting held on 08 April 2009. Thank you for attending to discuss the study.

Ethical opinion

1. The Research Ethics Committee (REC) noted that this was an offshoot of another study, which had already been granted ethics approval by this REC.

2. The REC queried whether it was expected that both parents would come along as they were concerned about the provision of questionnaires for single parents. Ms Soon said that in her experience that it was usually just the mother who showed up; if the father turned up too, then this was a bonus.

The REC queried whether if both parents came, then would the parents answer specific items in the questionnaires by discussion and mutual agreement or would they answer separately. Ms Soon said that maternal and paternal report was usually quite different.

3. The REC queried how Ms Soon knew how long to allow for the administration of the questionnaires. Ms Soon explained that she had used similar questions with other groups, and was therefore familiar with how long the questionnaires would take.

The members of the Committee present gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Ethical review of research sites

The Committee agreed that all sites in this study should be exempt from site-specific assessment (SSA). There is no need to submit the Site-Specific Information Form to any

Research Ethics Committee. The favourable opinion for the study applies to all sites involved in the research.

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission at NHS sites ("R&D approval") should be obtained from the relevant care organisation(s) in accordance with NHS research governance arrangements. Guidance on applying for NHS permission is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Approved documents

The documents reviewed and approved at the meeting were:

Document	*Version*	*Date*
Academic Supervisor	Dr Linda Franck	01 March 2009
Participant Consent Form: Assent Form, 8 - 16	2	02 March 2009
Participant Information Sheet: Teachers	1	09 March 2009
Participant Information Sheet: Parents	3	09 March 2009
Participant Information Sheet: Patients, 12 - 16	1	23 October 2008
Participant Information Sheet: Patients, 8 - 11	2	09 March 2009
Letter of invitation to participant	Parents, version 2	09 March 2009
Covering Letter		13 March 2009
Protocol		
Investigator CV	Ms Kristina Soon	
Application		16 March 2009
Participant Consent Form: Parent/ Guardian	2	09 March 2009
GP/Consultant Information Sheets	1	23 July 2007
Letter of invitation to participant	Teacher, version 1	09 March 2009
Interview Schedules/Topic Guides	Childr Attachment Interview, version 1	15 December 2008
Statistician Comments		13 March 2009
Questionnaire: Validated: SASA		
Questionnaire: Validated: Beck Youth Inventories Anxiety and Depression Scales		
Questionnaire: Validated: How People Feel About Me		
Questionnaire: Validated: ISS		
Questionnaire: Validated Questionnaire - SDQ: Self Report		
Questionnaire: Validated: Teacher, Social Skills		
Questionnaire: Validated: Teacher, Social Competence		

Questionnaire: Validated: How Important Are These Things to You		
Questionnaire: Validated: What Am I Like		
Questionnaire: Validated: SASC-R		
Questionnaire: Validated: Appearance Schemas Inventory		
Questionnaire: Validated: General Health Questionnaire		
Questionnaire: Validated: Social Skills, Parents		
Questionnaire: Validated: Social Competence, Parents		
Questionnaire: Validated: SDQ: Parent Report		
Questionnaire: Non-Validated: Background Information		
Questionnaire: Validated: PSI Item Booklet		
Questionnaire: Validated: DAS		
Questionnaire: Validated: SSQSR		
Questionnaire: Validated: SIAS		
Questionnaire: Validated: Feelings about my body		
Questionnaire: Non-validated: Questionnaire on Disfiguring Conditions	1	13 March 2009

Membership of the Committee

The members of the Ethics Committee who were present at the meeting are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Now that you have completed the application process please visit the National Research Ethics Website > After Review

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

We would also like to inform you that we consult regularly with stakeholders to improve our service. If you would like to join our Reference Group please email referencegroup@nres.npsa.nhs.uk.

09/H0713/19 **Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project

Yours sincerely

Enclosures:	List of names and professions of members who were present at the meeting and those who submitted written comments "After ethical review – guidance for researchers" SL-AR2 for other studies]
Copy to:	Dr Tracy Assari, Great Ormond Street Hospital/ Institute of Child Health R&D Department

Institute of Child Health/Great Ormond Street Hospital Research Ethics Committee

Attendance at Committee meeting on 08 April 2009

Committee Members:

Also in attendance:

Appendix G

Related Studies: Summary of Doctoral Studies Sharing Data Collection

INVOLVEMENT OF CO-RESEARCHERS

Four Doctorate in Clinical Psychology (DClinPsy) trainees have conducted research projects that overlap with this project. The reasons for conducting overlapping projects were as follows: 1) The current project is part of a larger programme of research which seeks to investigate a wide range of aspects of psychosocial functioning in the target population beyond those covered by the scope of the current study. 2) The Great Ormond Street Hospital paediatric dermatology service is the largest of its kind in the UK and the only one which has a clinical psychologist permanently on staff. As such, this department provides a unique research opportunity for psychologists interested in researching in this field. 3) The involvement of DClinPsy trainees, previously and currently, has progressed the research programme at a quicker pace, tested a broader range of research questions and significantly reduced workload, particularly in data collection. 4) Most importantly, by conducting several studies simultaneously, it was hoped that the number of times the same individuals are called upon to participate in this programme of research can be minimised.

Consultations were conducted with the Professorial Committee of the Research Department of Clinical, Educational and Health Psychology, UCL, the Research Directors of the UCL DClinPsy course, the Department of Postgraduate Studies at the Institute of Child Health, UCL, and the supervisors of this project, in order to agree the roles and responsibilities of each of the participating researchers. The UCL Research Department of Clinical, Educational and Health Psychology Professorial Committee Guidelines for shared research (see Appendix I) served as a framework for decision-making. The final project plan was agreed on the basis that it ensured that each student project would meet academic standards of originality,

independence and scope so that degree requirements were met by all student researchers involved.

There were four DClinPsy trainees who conducted research projects alongside the current project. The common theme of the projects was the investigation of factors that influence psychological adjustment in young people with disfiguring congenital dermatological conditions. As such, the participants were shared, the outcome variables were similar and overlapping, but the contributing variables measured were different. The measures used in each study are presented in Appendix H. The current author acted as field supervisor for all four projects as well as academic supervisor for two UCL DClinPsy students; Anna Seigal and Jenny Gibson. The research directors from each of the DClinPsy courses, as well as the supervisors of this project, have been consulted periodically to ensure that the projects have remained sufficiently independent.

The course affiliation, project titles and key hypotheses of each DClinPsy project are below:

UCL Doctorate in Clinical Psychology

Jennifer Gibson (JG)

Title: Do parental psychosocial experiences help to explain variation in the psychological functioning of children with congenital dermatological disfiguring conditions?

Key Hypotheses:

- Parents of congenitally disfigured young people will experience higher levels of psychological difficulty than parents in the normal population.
- Indices of parent and child psychosocial functioning will be associated.

- The relationship between parent and child psychological functioning will be moderated by child attachment style.

Khibza Hussain (KH)

Title: "Social skills, social competence and psychosocial adjustment in children with congenital dermatological disfiguring conditions".

Hypotheses:

- Social skills and social competence in disfigured adolescents will be significantly different to the normal adolescent population
- Social anxiety will be higher in disfigured adolescents than in the normal population.
- Social skills and social competence will be associated differentially with child psychosocial adjustment.

Anna Seigal (AS)

Title: "Exploring the relationship between attachment and psychological adjustment in children with congenital dermatological disfigurement"

Hypotheses:

- Children with disfigurements will have more psychological difficulties than children in the normal population.
- Children with congenital dermatological disfigurements will be more likely to be insecurely attached compared to the normal population.
- Attachment style will be associated with psychosocial adjustment.
- Children with facial disfigurements will be at greater risk of forming an insecure attachment than children with disfigurements affecting other areas of the body.
- The effect of the facial disfigurement on psychosocial adjustment will be mediated by attachment security.

Royal Holloway, University of London Doctorate in Clinical Psychology

Rebecca Mason (RM)

Title : Exploring the role of shame in the adjustment of adolescents with a congenital dermatological disfigurement.

Hypotheses:

- There will be a significant difference in anxiety, depression and self-perception between adolescents with congenital dermatological disfigurement, the non-disfigured clinical controls and school controls.
- Disfigured adolescents will experience significantly higher levels of internal, external and body shame than adolescents in the control groups.
- Shame will mediate the relationship with psychological adjustment in disfigured adolescents.

Appendix H

Related Studies: Table of All Measures Administered

Table H.1
Questionnaires in the Combined Data Collection by Researcher

	Researcher				
	KS	AS	JG	RM	KH
Child Measures				Adolescents Only	
	SDQ **SPP** **CAI** **OAS** **ISS**	SDQ SPP CAI	SDQ CAI	SDQ OAS ISS ESS	SDQ SPP SSQ SCQ SASA
Parent Measures					
	Dem. **Disfig** **SDQ**	Dem. Disfig SDQ	Dem. Disfig SDQ PSI-SF MHI-5 SIAS	Dem. Disfig SDQ	Dem. Disfig SDQ SSQ SCQ
Teacher Measures					
					SDQ SSQ SCQ

Researchers:
KS: Kristina Soon (current author)
AS: Anna Siegal
JG: Jennifer Gibson
RM Rebecca Mason
KH Khibza Hussain

Glossary:
SDQ: Strengths and Difficulties Questionnaire
SPP: Harter Self- Perception Profiles for Children and Adolescents
CAI: Child Attachment Interview
OAS: Other as Shamer Questionnaire
ISS: Internalised Shame Scale
ESS: Experience of Shame Scale
SASA: Social Anxiety Scale for Adolescents
SSQ: Social Skills Questionnaire
SCQ: Social Competence Questionnaire
Dem.: Demographic Questionnaire
Disfig: Disfiguremetn Questionnaire
SIAS: Social Interaction Anxiety Scale
PSI-SF: Parental Stress Index-Short Form
MHI-5: The Mental Health Inventory

Appendix I

Related Studies: UCL Research Department of Clinical, Educational and Health Psychology Professorial Committee Guidance on Shared Doctorates

Guidance to Undertaking a PhD while Supervising the Research of Professional Doctorate Students

1. There are many advantages to undertaking doctoral research as part of a collaborative team and this is encouraged. However in these circumstances it is essential that the contribution of each party and the way in which the thesis meets the following criteria (which apply to all doctoral programmes) is explicitly stated in a declaration and submitted with the thesis.

The thesis will make a distinct contribution to the knowledge of the subject and will afford evidence of originality as shown by the discovery of new facts and/or the exercise of independent critical power.

- In the case of Professional Doctorate students the declaration should be signed by each of the students involved in the project and their supervisor. The same examiner will be appointed for these theses.

- In the case of staff undertaking a PhD, the declaration should be signed by the staff member themselves and their supervisor and the declarations from all Professional Doctoral thesis based on data which overlaps at all with data reported in the PhD thesis should be submitted with the declaration. The examiners of the PhD thesis should be advised that these Professional Doctoral theses are available to them to consult at their request.

2. In planning their thesis work, team members should ensure that no studies are planned which involve completely overlapping data. For example Professional Doctorate student 1 might collect data on variables A, B and C in Year 1, Professional Doctorate student 2 might collect data on variables A, B and D in Year 2 and the staff member might analyse longitudinal data on the variables A and B in a PhD thesis study.

3. For the PhD upgrading the staff member should, in addition to the other documentation required, submit a draft of the declaration they envisage submitting with their thesis so that any questions that need to be resolved can be addressed at this stage and plans with the regard to use of shared data can be formally approved.

Appendix J

Related Studies: Declarations of Joint Working from the UCL Authors of the Related Studies

Anna Siegal

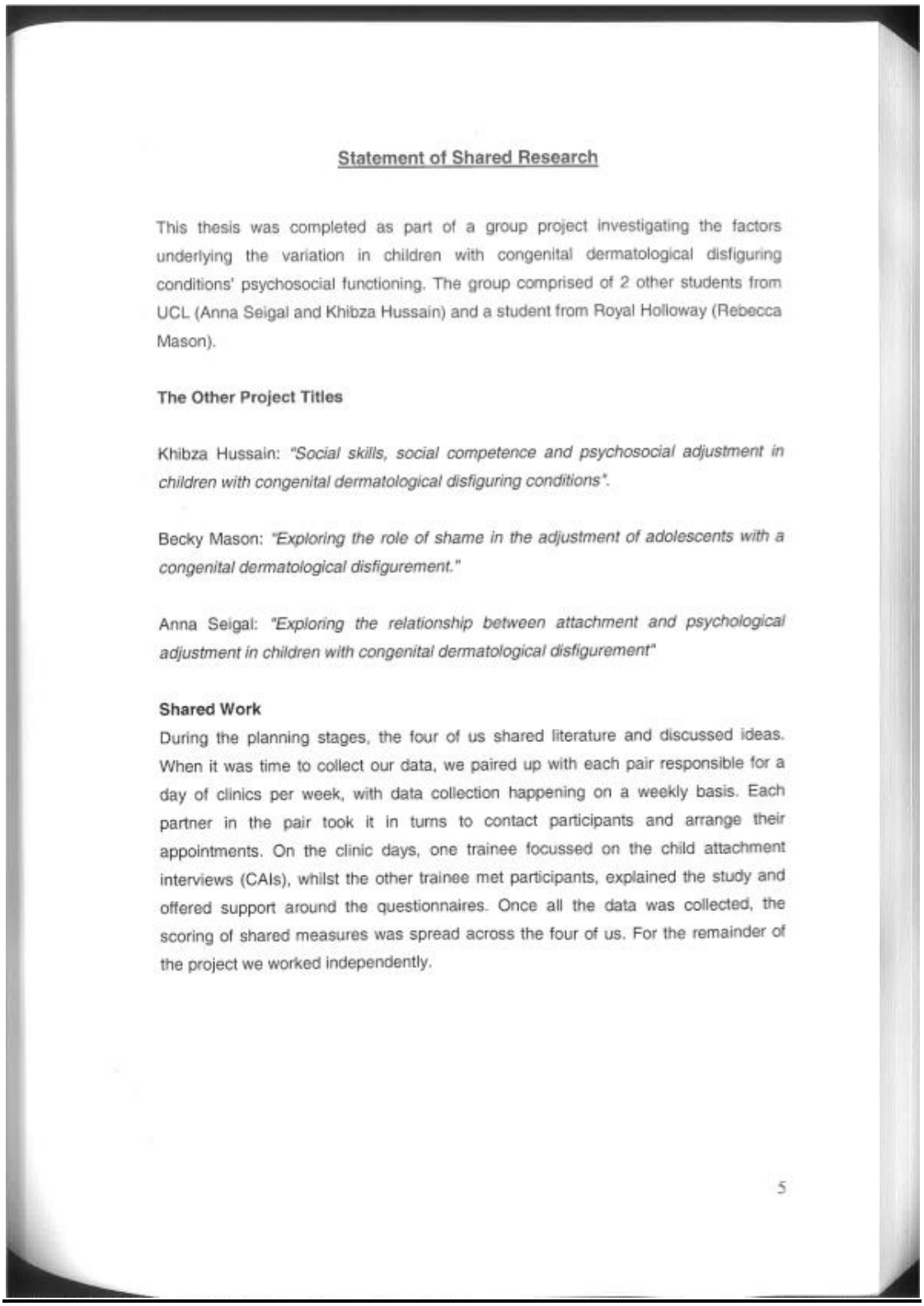
This study was part of a joint project, supervised by Kristina Soon. Two other trainees from UCL (Khibza Hussain and Jenny Gibson) and one trainee from Royal Holloway (Rebecca Mason) were involved. Data collection was shared equally between the four of us. The overall aim of the project was to explore psychological and social factors that contribute to positive adjustment in young people with congenital dermatological disfigurements. Each of us concentrated on a different aspect of this research, as outlined below.

My study is entitled "Exploring the relationship between attachment and psychological adjustment in children with congenital dermatological disfigurement" and explores the nature of the attachment relationship in children with congenital disfigurements and the relationship between attachment and psychological adjustment in this population.

Jenny Gibson's study is entitled "Does parent psychological functioning help explain variation in the psychosocial functioning of children with congenital dermatological disfiguring conditions?" It explores the psychological functioning of parents raising children with these conditions and the relationship between parent psychological functioning and child psychosocial functioning. It also investigates the role of child attachment as a moderating factor in the transfer of negative experiences between parents and their children.

Khibza Hussain's study is entitled "Social skills, social competence and psychosocial adjustment in children with congenital dermatological disfiguring conditions", and aims to explore the relative contributions of social skills and social competence on psychosocial adjustment.

Jennifer Gibson



Statement of Shared Research

This thesis was completed as part of a group project investigating the factors underlying the variation in children with congenital dermatological disfiguring conditions' psychosocial functioning. The group comprised of 2 other students from UCL (Anna Seigal and Khibza Hussain) and a student from Royal Holloway (Rebecca Mason).

The Other Project Titles

Khibza Hussain: *"Social skills, social competence and psychosocial adjustment in children with congenital dermatological disfiguring conditions".*

Becky Mason: *"Exploring the role of shame in the adjustment of adolescents with a congenital dermatological disfigurement."*

Anna Seigal: *"Exploring the relationship between attachment and psychological adjustment in children with congenital dermatological disfigurement"*

Shared Work

During the planning stages, the four of us shared literature and discussed ideas. When it was time to collect our data, we paired up with each pair responsible for a day of clinics per week, with data collection happening on a weekly basis. Each partner in the pair took it in turns to contact participants and arrange their appointments. On the clinic days, one trainee focussed on the child attachment interviews (CAIs), whilst the other trainee met participants, explained the study and offered support around the questionnaires. Once all the data was collected, the scoring of shared measures was spread across the four of us. For the remainder of the project we worked independently.

5

Khibza Hussain

This study was part of a joint project, supervised predominately by Kristina Soon (based at UCL and Great Ormond Street Hospital). Two other trainees from UCL (Anna Seigal and Jenny Gibson) and one trainee from Royal Holloway (Rebecca Mason) were involved. Data collection was shared equally between the four of us. The overall aim of the project was to explore psychological and social factors that contribute to positive adjustment in young people with congenital dermatological disfigurements. Each of us concentrated on a different aspect of this research, as outlined below.

My study is entitled "Social competence, social skills and psychosocial adjustment in adolescents with congenital dermatological disfiguring conditions" and explores the relative contributions of social skills and social competence on psychosocial adjustment.

Anna Seigal's study is entitled "Exploring the relationship between attachment and psychological adjustment in children with congenital dermatological disfigurement" and explores the nature of the attachment relationship in children with congenital disfigurements and the relationship between attachment and psychological adjustment in this population.

Jenny Gibson's study is entitled "Does parent psychological functioning help explain variation in the psychosocial functioning of children with congenital dermatological disfiguring conditions?" It explores the psychological functioning of parents raising children with these conditions and the relationship between parent psychological functioning and child psychosocial functioning. It also investigates the role of child attachment as a moderating factor in the transfer of negative experiences between parents and their children.

Rebecca Mason's study is entitled "Exploring the role of shame in the adjustment of adolescents with a congenital dermatological disfigurement" and explores the psychological profile of adolescents with a congenital dermatological disfigurement and to explore the role of shame in mediating this relationship.

Appendix K

Data Collection:
Demographics Questionnaire

BACKGROUND INFORMATION

(to be completed by a parent or guardian)

1) Child's age:

Age: …… years ……months

2**) Child's gender:**

☐ Male ☐ Female

3) School year:

☐ Year 3 ☐ Year 4 ☐

☐ Year 6 ☐ Year 7 ☐ Year 8

☐ Year 9 ☐ Year 10 ☐ Year 11

4) Occupation of main money earner:

Please tick **one box** to show which **best** describes the sort of work the main earner in your child's family does.

1 ☐

Modern professional occupations

Such as: teacher - nurse - physiotherapist - social worker - welfare officer - artist - musician - police officer (sergeant or above) - software designer

2 ☐

Clerical and intermediate occupations

Such as: secretary - personal assistant - clerical worker – office clerk - call centre agent - nursing auxiliary - nursery nurse

3 ☐

Senior managers or administrators

(Usually responsible for planning, organising and co-ordinating work and for finance) such as: finance manager - chief executive

4 ☐

Technical and craft occupations

Such as: motor mechanic - fitter - inspector - plumber - printer - tool maker - electrician - gardener - train driver

5 ☐

Semi-routine manual and service occupations

Such as: postal worker - machine operative - security guard - caretaker - farm worker - catering assistant - receptionist - sales assistant

6 ☐

Routine manual and service occupations

Such as: HGV driver - van driver - cleaner - porter - packer - sewing machinist - messenger - labourer - waiter / waitress - bar staff

7 ☐ **Middle or junior managers**
Such as: office manager - retail manager - bank manager - restaurant manager - warehouse manager - publican

8 ☐ **Traditional professional occupations**
Such as: accountant - solicitor - medical practitioner - scientist - civil / mechanical engineer

9 ☐ **Unemployed or on carer benefits**

5) How would you describe your child's ethnicity?
Please tick ONE box

A White
☐ British
☐ Any Other White background, *please write in*

B Mixed
☐ Any Mixed background, *please write in*

C Asian/Asian British
☐ Indian
☐ Pakistani
☐ Bangladeshi
☐ Chinese
☐ Any Other Asian background, *please write in*

D Black, Black British
☐ Caribbean
☐ African
☐ Any Other Black background, *please write in*

E Other ethnic group
☐ Any Other background, *please write in*

F Prefer not to say
☐

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Appendix L

Data Collection:
Medical Questionnaire

QUESTIONNAIRE ON DISFIGURING CONDITIONS

(to be completed by a parent or guardian)

1) What is the name of the condition that causes your child's disfigurement?

……

2) How old was your child when the disfigurement began?

☐ Present at birth ☐ Less than 2 years ☐ 2 years or older

3) On average, how often does your child attend hospital in a year?

☐ 1-2 times a year ☐ 3-5 times ☐ 6-10 times

☐ More than 10 times

4) Does this condition cause any disability that limits what your child can do, e.g. difficulty holding a pencil, difficulty seeing, difficulty walking or bending down?

☐ ☐ Yes No

If yes, please give brief details:

……

……

5) Does this condition cause any acute symptoms, intermittently, that can cause discomfort or distress, e.g. nasal congestion, breathlessness, seizures?

☐ ☐ Yes No

If yes, please give brief details:

……

……

6) Does this condition cause pain on a regular basis?

☐ ☐ Yes No

7) Does your child have any other medical or developmental problems?

☐ ☐ Yes No

If yes, please give brief details:

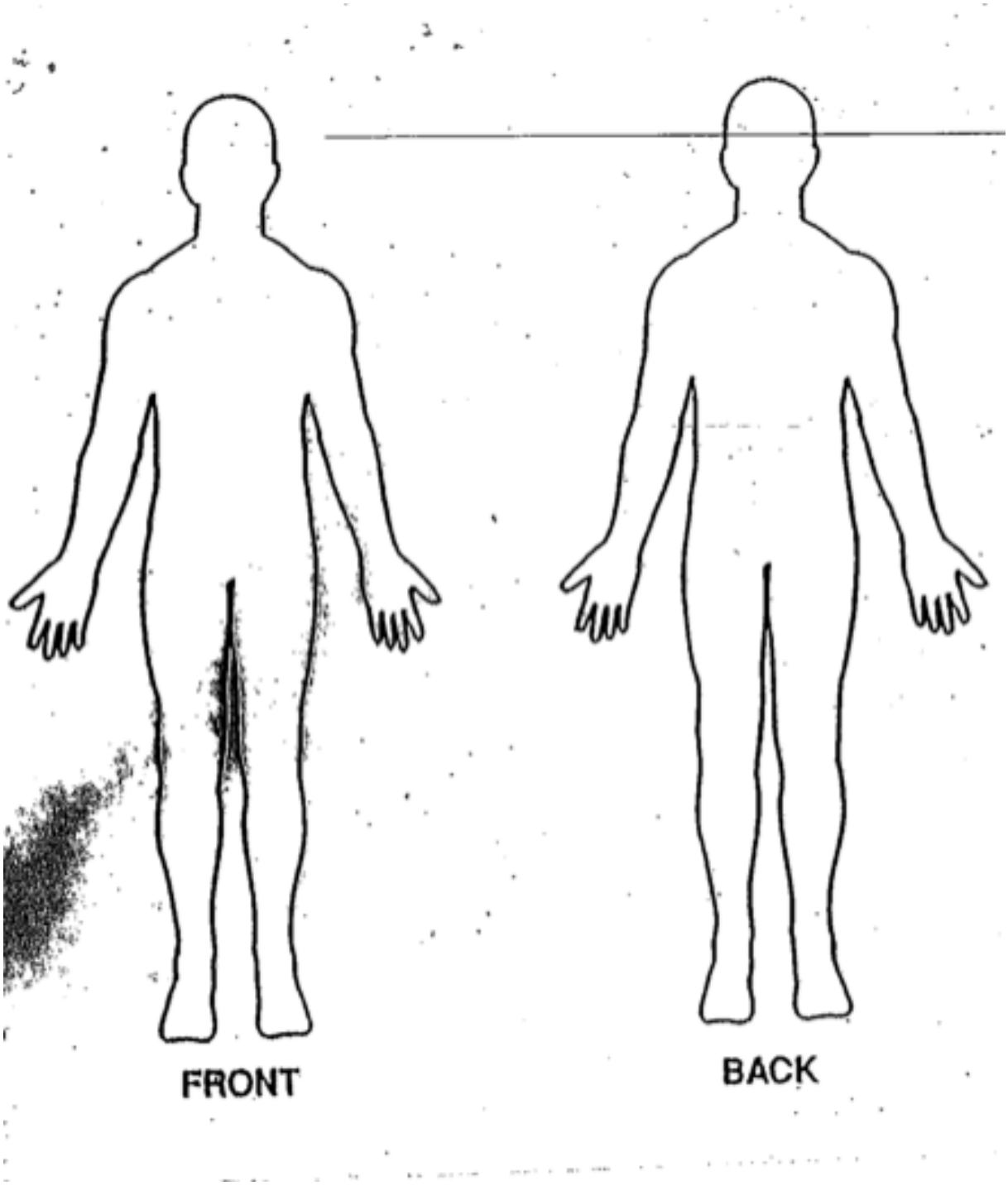
……

……

8) Could your child's medical condition(s) be life-threatening?

☐ ☐ Yes No

9) Please mark your child's disfigurement on the diagram below:



Appendix M

Strengths and Difficulties Questionnaire (SDQ)

i) parent report version
ii) child report version

M.i

Strengths and Difficulties Questionnaire P 4-16

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months.

Child's Name ..... Male/Female

Date of Birth.....

	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	☐	☐	☐
Restless, overactive, cannot stay still for long	☐	☐	☐
Often complains of headaches, stomach-aches or sickness	☐	☐	☐
Shares readily with other children (treats, toys, pencils etc.)	☐	☐	☐
Often has temper tantrums or hot tempers	☐	☐	☐
Rather solitary, tends to play alone	☐	☐	☐
Generally obedient, usually does what adults request	☐	☐	☐
Many worries, often seems worried	☐	☐	☐
Helpful if someone is hurt, upset or feeling ill	☐	☐	☐
Constantly fidgeting or squirming	☐	☐	☐
Has at least one good friend	☐	☐	☐
Often fights with other children or bullies them	☐	☐	☐
Often unhappy, down-hearted or tearful	☐	☐	☐
Generally liked by other children	☐	☐	☐
Easily distracted, concentration wanders	☐	☐	☐
Nervous or clingy in new situations, easily loses confidence	☐	☐	☐
Kind to younger children	☐	☐	☐
Often lies or cheats	☐	☐	☐
Picked on or bullied by other children	☐	☐	☐
Often volunteers to help others (parents, teachers, other children)	☐	☐	☐
Thinks things out before acting	☐	☐	☐
Steals from home, school or elsewhere	☐	☐	☐
Gets on better with adults than with other children	☐	☐	☐
Many fears, easily scared	☐	☐	☐
Sees tasks through to the end, good attention span	☐	☐	☐

Do you have any other comments or concerns?

Mii

Strengths and Difficulties Questionnaire

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of how things have been for you over the last six months.

Your Name ..... Male/Female

Date of Birth.....

	Not True	Somewhat True	Certainly True
I try to be nice to other people. I care about their feelings	☐	☐	☐
I am restless, I cannot stay still for long	☐	☐	☐
I get a lot of headaches, stomach-aches or sickness	☐	☐	☐
I usually share with others (food, games, pens etc.)	☐	☐	☐
I get very angry and often lose my temper	☐	☐	☐
I am usually on my own. I generally play alone or keep to myself	☐	☐	☐
I usually do as I am told	☐	☐	☐
I worry a lot	☐	☐	☐
I am helpful if someone is hurt, upset or feeling ill	☐	☐	☐
I am constantly fidgeting or squirming	☐	☐	☐
I have one good friend or more	☐	☐	☐
I fight a lot. I can make other people do what I want	☐	☐	☐
I am often unhappy, down-hearted or tearful	☐	☐	☐
Other people my age generally like me	☐	☐	☐
I am easily distracted, I find it difficult to concentrate	☐	☐	☐
I am nervous in new situations. I easily lose confidence	☐	☐	☐
I am kind to younger children	☐	☐	☐
I am often accused of lying or cheating	☐	☐	☐
Other children or young people pick on me or bully me	☐	☐	☐
I often volunteer to help others (parents, teachers, children)	☐	☐	☐
I think before I do things	☐	☐	☐
I take things that are not mine from home, school or elsewhere	☐	☐	☐
I get on better with adults than with people my own age	☐	☐	☐
I have many fears, I am easily scared	☐	☐	☐
I finish the work I'm doing. My attention is good	☐	☐	☐

Your signature ..... Today's date .....

Thank you very much for your help

Appendix N

Harter Self-Perception Profile (SPP)

i) Child version (SPPC)
ii) Adolescent version (SPPA)

Appendix N.i

What I Am Like

SAMPLE SENTENCE

	Really True for me	Sort of True for me				Sort of True for me	Really True for me
(a)	☐	☐	Some kids would rather play outdoors in their spare time	BUT	Other kids would rather watch T.V.	☐	☐
1.	☐	☐	Some kids feel that they are very good at their school work	BUT	Other kids *worry* about whether they can do the school work assigned to them.	☐	☐
2.	☐	☐	Some kids find it *hard* to make friends	BUT	Other kids find it's pretty *easy* to make friends.	☐	☐
3.	☐	☐	Some kids do *very well* at all kinds of sports	BUT	Other kids *don't* feel that they are very good when it comes to sports.	☐	☐
4.	☐	☐	Some kids are *happy* with the way they look	BUT	Other kids are *not* happy with the way they look.	☐	☐
5.	☐	☐	Some kids often do not like the way they behave	BUT	Other kids usually *like* the way they behave.	☐	☐
6.	☐	☐	Some kids are often *unhappy* with themselves	BUT	Other kids are pretty *pleased* with themselves.	☐	☐
7.	☐	☐	Some kids feel like they are *just as smart* as other kids their age	BUT	Other kids aren't so sure and *wonder* if they are as smart.	☐	☐
8.	☐	☐	Some kids have *a lot* of friends	BUT	Other kids *don't* have very many friends.	☐	☐

	Really True for me	Sort of True for me				Sort of True for me	Really True for me
9.	☐	☐	Some kids wish they could be a lot better at sports	BUT	Other kids feel they are good enough at sports.	☐	☐
10.	☐	☐	Some kids are *happy* with their height and weight	BUT	Other kids wish their height or weight were *different*.	☐	☐
11.	☐	☐	Some kids usually do the *right* thing	BUT	Other kids often *don't* do the right thing.	☐	☐
12.	☐	☐	Some kids *don't* like the way they are leading their life	BUT	Other kids *do* like the way they are leading their life.	☐	☐
13.	☐	☐	Some kids are pretty *slow* in finishing their school work	BUT	Other kids can do their school work *quickly*.	☐	☐
14.	☐	☐	Some kids would like to have a lot more friends	BUT	Other kids have as many friends as they want.	☐	☐
15.	☐	☐	Some kids think they could do well at just about any new sports activity they haven't tried before	BUT	Other kids are afraid they might *not* do well at sports they haven't ever tried.	☐	☐
16.	☐	☐	Some kids wish their body was *different*	BUT	Other kids *like* their body the way it is.	☐	☐
17.	☐	☐	Some kids usually *act* the way they know they are *supposed* to	BUT	Other kids often *don't* act the way they are supposed to.	☐	☐
18.	☐	☐	Some kids are *happy* with themselves as a person	BUT	Other kids are often *not* happy with themselves.	☐	☐
19.	☐	☐	Some kids often *forget* what they learn	BUT	Other kids can remember things *easily*.	☐	☐
20.	☐	☐	Some kids are always doing things with *a lot* of kids	BUT	Other kids usually do things by *themselves*.	☐	☐

	Really True for me	Sort of True for me				Sort of True for me	Really True for me
21.	☐	☐	Some kids feel that they are *better* than others their age at sports	BUT	Other kids *don't* feel they can play as well	☐	☐
22.	☐	☐	Some kids wish their physical appearance (how they look) was *different*	BUT	Other kids *like* their physical appearance the way it is.	☐	☐
23.	☐	☐	Some kids usually get in *trouble* because of things they do	BUT	Other kids usually *don't* do things that get them in trouble.	☐	☐
24.	☐	☐	Some kids like the kind of *person* they are	BUT	Other kids often wish they were someone else.	☐	☐
25.	☐	☐	Some kids do *very well* at their classwork	BUT	Other kids *don't* do very well at their classwork.	☐	☐
26.	☐	☐	Some kids wish that more people their age liked them	BUT	Other kids feel that most people their age *do* like them.	☐	☐
27.	☐	☐	In games and sports some kids usually *watch* instead of play	BUT	Other kids usually *play* rather than just watch.	☐	☐
28.	☐	☐	Some kids wish something about their face or hair looked *different*	BUT	Other kids *like* their face and hair the way they are.	☐	☐
29.	☐	☐	Some kids do things they know they *shouldn't* do	BUT	Other kids *hardly ever* do things they know they shouldn't do.	☐	☐
30.	☐	☐	Some kids are very *happy* being the way they are	BUT	Other kids wish they were *different*.	☐	☐
31.	☐	☐	Some kids have trouble figuring out the answers in school	BUT	Other kids almost *always* can figure out the answers.	☐	☐
32.	☐	☐	Some kids are popular with others their age	BUT	Other kids are *not* very popular.	☐	☐

	Really True for me	Sort of True for me				Sort of True for me	Really True for me
33.	☐	☐	Some kids *don't* do well at new outdoor games	BUT	Other kids are *good* at new games right away.	☐	☐
34.	☐	☐	Some kids think that they are good looking	BUT	Other kids think that they are not very good looking	☐	☐
35.	☐	☐	Some kids behave themselves very well	BUT	Other kids often find it hard to behave themselves.	☐	☐
36.	☐	☐	Some kids *are* not very happy with the way they do a lot of things	BUT	Other kids think the way they do things is *fine*.	☐	☐

Appendix N.ii

What I Am Like

	Really True for me	Sort of True for me	SAMPLE SENTENCE			Sort of True for me	Really True for me
(a)	☐	☐	Some teenagers like to go to movies in their spare time	BUT	Other teenagers would rather go to sports events.	☐	☐
1.	☐	☐	Some teenagers feel that they are just as smart as others their age	BUT	Other teenagers aren't so sure and wonder if they are as smart.	☐	☐
2.	☐	☐	Some teenagers find it hard to make friends	BUT	For other teenagers it's pretty easy.	☐	☐
3.	☐	☐	Some teenagers do very well at all kinds of sports	BUT	Other teenagers don't feel that they are very good when it comes to sports.	☐	☐
4.	☐	☐	Some teenagers are *not* happy with the way they look	BUT	Other teenagers *are* happy with the way they look.	☐	☐
5.	☐	☐	Some teenagers feel that they are ready to do well at a a part-time job	BUT	Other teenagers feel that they are not quite ready to handle a part-time job.	☐	☐
6.	☐	☐	Some teenagers feel that if they are romantically interested in someone, that person will like them back	BUT	Other teenagers worry that when they like someone romantically, that person *won't* like them back.	☐	☐
7.	☐	☐	Some teenagers usually do the right thing	BUT	Other teenagers often don't do what they know is right.	☐	☐
8.	☐	☐	Some teenagers are able to make really close friends	BUT	Other teenagers find it hard to make really close friends.	☐	☐
9.	☐	☐	Some teenagers are often disappointed with them-selves	BUT	Other teenagers are pretty pleased with themselves.	☐	☐
10.	☐	☐	Some teenagers are pretty slow in finishing their school work	BUT	Other teenagers can do their school work more quickly.	☐	☐
11.	☐	☐	Some teenagers have a lot of friends	BUT	Other teenagers don't have very many friends.	☐	☐
12.	☐	☐	Some teenagers think they could do well at just about any new athletic activity	BUT	Other teenagers are afraid they might not do well at a new athletic activity.	☐	☐

1

	Really True for me	Sort of True for me				Sort of True for me	Really True for me
13.	☐	☐	Some teenagers wish their body was different	BUT	Other teenagers like their body the way it is.	☐	☐
14.	☐	☐	Some teenagers feel that they *don't* have enough skills to do well at a job	BUT	Other teenagers feel that they *do* have enough skills to do a job well.	☐	☐
15.	☐	☐	Some teenagers are *not* dating the people they are really attracted to	BUT	Other teenagers *are* dating those people they are attracted to.	☐	☐
16.	☐	☐	Some teenagers often get in trouble for the things they do	BUT	Other teenagers usually *don't* do things that get them in trouble.	☐	☐
17.	☐	☐	Some teenagers do have a close friend they can share secrets with	BUT	Other teenagers do not have a really close friend they can share secrets with.	☐	☐
18.	☐	☐	Some teenagers don't like the way they are leading their life	BUT	Other teenagers do like the way they are leading their life.	☐	☐
19.	☐	☐	Some teenagers do very well at their classwork.	BUT	Other teenagers don't do very well at their classwork.	☐	☐
20.	☐	☐	Some teenagers are very hard to like	BUT	Other teenagers are really easy to like.	☐	☐
21.	☐	☐	Some teenagers feel that they are better than others their age at sports	BUT	Other teenagers *don't* feel they can play as well.	☐	☐
22.	☐	☐	Some teenagers wish their physical appearence was different	BUT	Other teenagers like their physical appearence the way it is.	☐	☐
23.	☐	☐	Some teenagers feel they are old enough to get and keep a paying job	BUT	Other teenagers do not feel they are old enough, yet, to really handle a job well.	☐	☐
24.	☐	☐	Some teenagers feel that people their age will be romantically attracted to them.	BUT	Other teenagers worry about whether people their age will be attracted to them.	☐	☐
25.	☐	☐	Some teenagers feel really good about the way they act	BUT	Other teenagers *don't* feel that good about the way they often act.	☐	☐
26.	☐	☐	Some teenagers wish they had a really close friend to share things with	BUT	Other teenagers *do* have a close friend to share things with.	☐	☐
27.	☐	☐	Some teenagers are happy with themselves most of the time	BUT	Other teenagers are often not happy with themselves.	☐	☐
28.	☐	☐	Some teenagers have trouble figuring out the answers in school	BUT	Other teenagers almost always can figure out the answers.	☐	☐

	Really True for me	Sort of True for me				Sort of True for me	Really True for me
29.	☐	☐	Some teenagers are popular with others their own age	BUT	Other teenagers are not very popular.	☐	☐
30.	☐	☐	Some teenagers don't do well at new outdoor games	BUT	Other teenagers are good at new games right away.	☐	☐
31.	☐	☐	Some teenagers think that they are good looking	BUT	Other teenagers think that they are not very good looking.	☐	☐
32.	☐	☐	Some teenagers feel like they could do better at work they do for pay	BUT	Other teenagers think that they are doing really well at work they do for pay.	☐	☐
33.	☐	☐	Some teenagers feel that they are fun and interesting on a date	BUT	Other teenagers wonder about how fun and interesting they are on a date.	☐	☐
34.	☐	☐	Some teenagers do things they know they shouldn't do	BUT	Other teenagers hardly ever do things they know they shouldn't do.	☐	☐
35.	☐	☐	Some teenagers find it hard to make friends they can really trust	BUT	Other teenagers *are* able to make close friends they can really trust.	☐	☐
36.	☐	☐	Some teenagers like the kind of person they are.	BUT	Other teenagers often wish they were someone else.	☐	☐
37.	☐	☐	Some teenagers feel that they are pretty intelligent	BUT	Other teenagers question whether they are intelligent.	☐	☐
38.	☐	☐	Some teenagers feel that they are socially accepted	BUT	Other teenagers wished that more people their age accepted them.	☐	☐
39.	☐	☐	Some teenagers do not feel that they are very athletic	BUT	Other teenagers feel that they are very athletic.	☐	☐
40.	☐	☐	Some teenagers really like their looks	BUT	Other teenagers wished they looked different.	☐	☐
41.	☐	☐	Some teenagers feel that they are really able to handle the work at a paying job	BUT	Other teenagers wonder if they are really doing as good a job at work as they should be doing.	☐	☐
42.	☐	☐	Some teenagers usually *don't* go out with the people they would really like to date	BUT	Other teenagers *do* go out with the people they really want to date.	☐	☐
43.	☐	☐	Some teenagers usually act the way they know they are supposed to	BUT	Other teenagers often don't act the way they are supposed to.	☐	☐
44.	☐	☐	Some teenagers *don't* have a friend that is close enough to share really personal thoughts with	BUT	Other teenagers do have a close friend that they can share personal thoughts and feelings with.	☐	☐
45.	☐	☐	Some teenagers are very happy being the way they are	BUT	Other teenagers wish they were different.	☐	☐

Appendix O

Child Attachment Interview (CAI): Adapted Interview

Child Attachment Interview - adapted
(Target, Fonagy, Schmueli-Goetz, 2003)

"This is an interview about you and your family. *(Here you can ask whether the child knows what an interview is, and make it clear that you want to know about his/her own point of view about things)*. For each question I will ask you to give me some examples. This interview is not a test and there are no right or wrong answers. I would just like you to tell me what you and your family are like from your point of view. The interview will last about half an hour maybe a bit more."

1) Can you tell me about the people in your family; the people living together in your house? Anyone else?

2) (Warm up question). **Tell me 3 words that describe yourself, not what you look like, but what sort of person you are.**
Prompts: Some children may find it helpful to imagine writing a letter to a pen pal or you could prompt by asking how someone who knows him/her might describe him/her

3) Tell me 3 words to describe your relationship with your mum; what it's like to be with your Mum?

4) What happens when your Mum gets cross with you or tells you off?
Prompts: If you've done something wrong or done something to make her cross with you, what does she usually say or do? How do you feel when that happens? How do you think your mum feels when that happens? Why do you think she does….? (Insert whatever the child says the mother does)

5) Can you tell me 3 words to describe your relationship with your Dad; what it's like to be with your Dad?

6) What happens when your Dad gets cross with you or tells you off?

At this point, ask same questions about 3rd and 4th caregivers if you have decided to include someone as an additional attachment figure.

7) Can you tell me about a time when you were upset and wanted help?
Prompt: What happened? What did your Mum/Dad do?

8) Do you ever feel that your parents don't really love you?

9) What happens if someone is mean to you about the way that you look?

10) Do your parents sometimes argue?
Prompts: How do you feel? Why do you feel like that? Why do you think they do that? How do you think they feel? Do they know how you feel?

11) a. In what way would you like to be like/not like your Mum?
b. In what ways would you like to be like/not like your Dad?

12) (Closure Question) **If you could have 3 wishes, what would they be?**

End the interview by thanking the child and saying how helpful they have been and that you know that some of the questions are difficult and you really appreciate their help.

V1.15.12.08

Appendix P

Child Attachment Interview (CAI): Anonymised sample transcript

CAI Coding Table

Date of Rating: **Name of Rater:** **KS** **Child ID:** **122** **Date of Testing:**

Background Information **Lives with mum, dad and puppy.**

Questions	Transcript of Episodic Examples / General notes	Notes/Relevant Scales
Self – 3 ways to describe yourself		
Funny	I had my friend round and I was making her laugh with jokes and like remembering times when we were together (and what was funny about that?) ooo I don't know she just laughs for no reason (does she?) yeah and it makes me laugh	
Kind	There was one… o yeah. Erm well, my next door neighbour, well in front of us, she has a little girl and shes like 4 and I gave up like two of my toys, like, for her and made her a necklace.	
Energetic	Well I always go to bed very late because I like to be excited with my puppy …and yeah I do a lot of running because I go to cross-countries and I got medals.	
3 ways describe relationship with Mum		
Fighting	It was like an argument basically, not an actual fight. Erm it was a very strange time, we started fighting because I gave my puppy the wrong type of food and then she got angry with herself because we bought her this food stuff and it was bad for her and I was trying to help her but she got angry with me.	
Loveable	Well she's very nice and she always buys me things, which I very like and yeah, and we would hug and things. (Can you tell me about one particular time when it was loveable being with your mum?) O Yeah I was angry because I had to change my clothes but I didn't want to because I was very comfy in them and I was very sad and we went into a shop and she'd buy me a sweeties and yeah… she did something else but yeah.	
Gentle	…I had fallen over and it really hurt because we were in Peru and I fell over and scraped my knee on the wooden stairs and she slept with me at night because I was very, she was very kind to me, kind and gentle.	
Happens when Mum is cross	I was quite small and then she told me that she was crying at night, because I was small and I cut my eyelashes because I was small and I didn't know what to do and I was very sad because I found out that was my mum was crying (so what did she say or do to you at the time?) o yeah, she erm, …what did she do? She shouted at me and just shouted at me really but she didn't do anything really (OK, and what sort of things was she shouting at you?) Like kind of giving me a lesson, about like, but in a shouty way, like why did you do that, you're not suppose to do that, that's very dangerous…. (How did you feel when your mum was doing that shouty thing?) I felt very, what's it called, guilty and a bit horrible and hurt (why did you feel that way?) because I realised what I've done and I realised that I made my mum angry, so I didn't like it. (How do you think your mum felt?) Very worried because I could have cut it too short and it could have started bleeding or something. (Why do you think she got so angry about that? Why was she telling you off?)	

	Because she worried and she didn't want anything to happen to me because she loves me. (So do you think it was fair that your mum got cross with you like that?) Yeah (whys that?) Because I wasn't really thinking at the time, so, I was kind of like bored so, she had the right to do it to tell me off to help me realise what I had done. (So how did it all finish then?) I said sorry (What did your mum say or do?) I forgive you but don't do it again.	
3 ways describe relationship with Dad		
Hardly ever (never) fight	Well it's always, he's a very kind person, he has a lot a patience. My mum always wishes that she had as much patience as my dad. He's very nice. (So he's very patient?) Yeah, with me and like with the family.	
Smooth	Smooth is like another word for gentle – everything runs smoothing and it's all calm. (So tell me about s time when it was just like that with you and your dad) Well on the train here I felt very sick, and he helped me and played a game with me – it helped me.	
Loveable	We went to a centre park and me and my dad were doing laser gun shooting with my cousin but it was really fun and we were helping each other and it was a very nice time to be together. (What was so nice about it?) It was something that we both enjoy and you can work as a team.	
Happens when Dad is cross	He sometime gets my mum involved so my mum actually does most of the telling off but my dad does – well he doesn't really shout at me, he just, because I don't like my dad getting cross because he hardly ever does it makes me really sad so he just has to say that's very naughty and I feel sad. (Do you remember the last time he did that?) I think I did – well there was one time which was, I was again small but not that small and we used to have a cat – a kitten – and she was on the sofa and I want to get her out so I pulled her leg or her tail or something and my dad saw me and he got very cross because it wasn't very nice. (What did you dad say or do?) He didn't let me go near her for about, not a long time, like 10 minutes or half and hour or something. (Did he say anything to you?) Just, yeah, he said, that was not nice and you shouldn't do that because it will hurt her. (How did he say that to you? …A bit shouty or?) Serious. (How did you feel when your dad said that to you?) Tearful and guilty because, and sad and angry with myself. (Why did you feel all those feelings do you think?) Because my dad never gets cross and I don't really like it when he does and I felt, I realised what it must have felt like for my cat being hurt so I didn't really like the feeling. (How do you think your dad felt when he was telling you you shouldn't hurt the cat?) A bit disappointed in me because and I nearly always behave well so when sees that I do something bad he doesn't really. (Why do you thin he got cross at you at that time and told you?) Because he knows that I never do that kind of things so he saw me do it and was surprised and he was very angry. (Why was he so angry do you think?) Because it's not very nice to hurt animals, because they're not like humans they cant tell you what there thinking. (Do you think your dad knew how you felt when he was telling you off?) A bit – well because adults don't really understand children – not all of the time. So he probably thought that I felt that I didn't understand, that I didn't care, but I do care. (Do you think it was fair that he told you off?) 'Nods' (Why is that?) Because I shouldn't have done that and it was a very bad thing (So how did it end?) well I – well he sent me to my room because I couldn't be near her and I said sorry to him….he said now you have to apologise to, so I said sorry and he gave me a hug and I apologised to my cat.	
Child upset	Because I have two main friends and they started calling me 'annoying' and 'horrible' and 'selfish' and – but I wasn't I didn't do anything so I don't know why she ever said that and I didn't know where all the rest of my friends – my other friends were – so I was very sad and they were all angry with me so they went away and left and I had no one to play with and I was very hurt. (So how did you feel when all that was happening?) I felt worried that I wouldn't have any more friends, I don't know why, and I felt horrible inside that they said that to me (How do you think they were felling, or you	

	friend was feeling when she was saying all those things to you?) I don't think they cared, they just said it because they said it, no reason (Why did they say all those things to you do you think?) I have no idea what we were talking about – I was just playing but they found it annoying because I was playing with them – not with them but singing, not singing but like… and they found it very annoying so they just said your so annoying, your so horrible, I don't like you and they just went away and left me. (Do you thin that they knew how you felt when they said that to you?) No. (So what did you do?) Eventually I found more friends, I found my other friends and I went to play with them and I told them how I felt and they did understand and they helped me and they went to talk to the other people who said that to me. And then they said sorry and now were all friends again. (So did you tell your mum and dad what happened?) yes (and what did they say or do?) Well they found it very horrible because one of the girls that said that to me is always horrible and mean to everybody…but sometimes she can be nice when she wants to so that's why I was her friend but she can be horrible. So they probably felt like 'o that's not very nice to do that to my daughter, I wish she would play with somebody else'. (So how did you feel when you told your mum and dad what happened?) Well I didn't really feel anything, I just felt proud that I told them, I just felt happy that I could tell them (Were you pleased, glad that told them?) they seem pleased. (Do you think it was good idea to tell your friends and your mum and dad what happened?) Yeah because if I didn't then I would just, I would have like no friends, I'd have friends but I wouldn't be friends with the people that I would like to be friends with and I would just be sad and never be friends again with them.	
Feel parents love	No. Because even if they're angry with me I do know that they love me	
Happens when someone's mean	(Tel me of a time when someone was mean about the way you look?) Well she wasn't very mean. We went to a park with a friend, well somebody I didn't know and a friend from school – we were just playing and getting ready to go. A girl younger then me about 7, was just there said to me, 'you do realise you've got a purple lip', I said 'yes its been there all my life' and she said 'its really big and horrible and does not suit you' and I was just like ' what' because that's not very nice and me and my friend just said 'excuse me that's not very nice and me and my friend just went because we don't want to stay with somebody's so horrible. (How did you feel?) Hurt, angry and sad and worried as well (Why?) because I thought if that's what she felt maybe other people feel like that as well, maybe everybody felt like that but just didn't want to say it. (How do you think she felt when she was saying all this stuff to you?) Didn't care, just said it because. (Why did she say it to you?) Why would you want to tell somebody because you should just keep tit yourself it might hurt there feelings. I don't know really why she said it. (What did you do?) Told my friends mum, to tell her and she just stood there looking at her, she wanted to go talk to her and say why did you say that to my daughter friends but my friend said no and we just went. I kept looking back she was just staring at me. (did you tell your mum and dad what happened?) Yes. (What did they say or do?) they were just surprised because its not very nice and they felt like…they were pleased with me that I told them…and a bit sad for me because its not very nice. (did you think it was good idea to your mum and dad? Why?) Yeah, because if I just keep it to myself all those times, all those years would be gone and if I was to meet someone else like you, they would say as well is there something horrible that hidden and if I told them, my mum and dad wouldn't know what I was talking about so couldn't help the doctor understand, so they have to know. (Is it helpful talking you mum and dad about things like that?) Yes.	
Somebody you care about, not here anymore	My Cousin died but they live in Peru and we only go there once every two years, I did feel sad but I didn't know him but I didn't see him a lot. He had a heart attack. My grandpa died but I hadn't even seen him because he died when my mum was 20 – I wasn't even born. (How did you find out your cousin died? How old were you?) I think it was last year… or two years ago. My mum told me I think. (How did you feel?) Shocked that something had happened like that because	

	nobody in my life had died while I'm born – shocked and very sad to hear that bad news. (How did your mum and dad feel?) I don't really know, because its not my dads family but he probably still felt very sad and my mum, it was her nephew, she was crying I think. When we went next time to Peru, we went to the grave. We went to see them and everybody was crying. (why were your mum and dad feeling like that?) because they knew him very well and it was nice having him around , he was kind and for them to go out of your life and not see them anymore…upsetting. (Did your mum and dad know how you felt?) Don't thinks so. (Did they do anything to try to make you feel better?) No because I didn't cry or anything I was just shocked. I was fine.	
Away from parents	No but I will next year – a whole week, a bit scared but its going to be fun. I have stayed a whole day and a whole night and then the next day I stayed another whole day and night but I didn't actually see them and I just called them. It was a t my grandmas house, so very close. So if anything happened they can just come… (How did you feel when you stayed away from your mum and dad?) The last night I did cry because I missed them. It was like in the middle of the night so I couldn't do anything about it but I woke up and my cousin read me a book. (How do you think your mum and dad felt?) They might have felt worried because they are protective over me. But they knew I was in safe hands because my grandma looks after me. (DO you think your mum and dad knew that you might be missing them a bit?) Yes. Because they know that I love them and I think they are very nice. (What was it like when you got to see them again?) I was very happy and when my mum opens the dad she always runs and hugs me. (How do you think your mum and felt when they saw you again for the first time?) Very happy, they were asking me questions, like how was it, did you have a nice time?	
Parents argue	Sometimes, not like a proper argument. Sometimes my dad does the shopping and he buys the wrong thing and its something important my mum gets quite annoyed. (What does she say or do?) She'll say ' o why did you do that, o dear….' (and what does you dad say or do?) he says ' I didn't know' and just goes out and buys the right thing. (How do you feel when you hear your mum and dad arguing?) I don't really like it because I want them to be happy, be a happy family. (How do you think your mum and dad are feeling when they are having that argument?) well my mum would probably feel angry with my dad and annoyed and a bit stressed out and my dad would probably feel shocked and weird. (Do you think they know how you feel when they argue?) Sometimes I don't think they notice that I'm listening or that I know that they are arguing. But if I am around, near them when they argue, I don't think they'll even notice me. (Why do you think they argue about things like that?) I have no idea. Just the kind of people they are.	
Ways like Mum	She can do things like cooking, cleaning…I wouldn't really like to do cleaning but cooking and organising. I do try to be organised but sometimes I just cant manage it so I would like to be more organised and do cooking and get some things she can do	
Ways not like Mum	Stressed out, inpatient	
Ways like Dad	More patient then I can't get angry because he never gets angry.	
Ways not like Dad	Not Organised	
3 wishes	1. Not to have this birth mark 2. Mum could be more patient 3. More friends at school	

Appendix Q

Child Attachment Interview (CAI): Coding Form

CAI RATING FORM

PARTICIPANT NUMBER:
RATER:

Emotional Openness

Balance of positive and Negative references to attachment figures

Use of examples

	Mum	Dad
Preoccupied Anger		
Idealisation		
Dismissal/Derogation		
Resolution of Conflict		
Disorganisation		
Overall coherence		

	Classification	Sub-classification
Mum		
Dad		

Appendix R

Measure of External Shame:
Other as Shamer Questionnaire (OAS)

How other people feel about me.

(Goss, Gilbert & Allan, 1994)

We all try to guess what other people think or feel about us. Do they like me? Do they think I'm smart or good-looking?
Below are thoughts that you might have about what other people are thinking or feeling about you.
Circle the number that is closest to how often you think each one.

Never	Almost never	Sometimes	Often	Always
1	2	3	4	5

1. Other people think I'm not as good as them	1	2	3	4	5
2. I think other people respect me	1	2	3	4	5
3. I think other people look down on me	1	2	3	4	5
4. I feel other people think I'm not good enough	1	2	3	4	5
5. Other people like being around me	1	2	3	4	5
6. Other people see me as small and meaningless	1	2	3	4	5
7. I feel nervous about what other people think of me	1	2	3	4	5
8. Other people show an interest in me	1	2	3	4	5
9. People see me as unimportant compared to others	1	2	3	4	5
10. Other people think there's something wrong with me	1	2	3	4	5
11. I know that other people like me	1	2	3	4	5
12. Other people think there's something missing in me	1	2	3	4	5
13. Other people see me as empty and unfulfilled	1	2	3	4	5
14. Other people often praise me for things I do well	1	2	3	4	5
15. Other people like having me around them	1	2	3	4	5
16. Other people think I don't have control of my body or my feelings	1	2	3	4	5
17. Others see me as being weak	1	2	3	4	5
18. Others criticise me when I make a mistake	1	2	3	4	5
19. Other people see me as a strong person	1	2	3	4	5
20. Other people always remember my mistakes	1	2	3	4	5

21. People try to stay away from me	1	2	3	4	5
22. I think people see my good qualities	1	2	3	4	5
23. People look for my faults	1	2	3	4	5
24. Other people put me down a lot	1	2	3	4	5
25. I think other people can see all the things that are wrong with me	1	2	3	4	5

Appendix S

Measure of Internal Shame: Internalised Shame Scale (ISS)

ISS

Below is a list of sentences about feelings or thoughts that you might have now and then. Most of these feelings and thoughts are sad or upsetting in some way. Some people have these feelings a lot, while others only have them a little. But we all have them sometimes.

Try to answer as honestly as you can.

Read each sentence carefully and circle the number on the right side that is closest to how often you feel what the sentence says. Make sure that you answer all of them.

	Never	Rarely	Sometimes	Often	Almost Always
1. I feel like I am never quite good enough	0	1	2	3	4
2. I feel left out	0	1	2	3	4
3. I think that people look down on me	0	1	2	3	4
4. Overall, I feel that I am a success	0	1	2	3	4
5. I get cross with myself and put myself down	0	1	2	3	4
6. I worry that other people think bad things about me	0	1	2	3	4
7. I feel that I'm not as good as other people	0	1	2	3	4
8. I see myself as being very small and unimportant	0	1	2	3	4
9. I feel I have a lot to be proud of	0	1	2	3	4
10. I worry that I'm not good enough	0	1	2	3	4
11. I feel like there's something basically wrong with me	0	1	2	3	4
12. When I compare myself to others I am just not as important	0	1	2	3	4
13. I worry that other people will find out about all the things that are wrong with me	0	1	2	3	4
14. I feel that I have a number of good qualities	0	1	2	3	4
15. I try to be perfect but I always fail	0	1	2	3	4

	Never	Rarely	Sometimes	Often	Almost Always
16. I think other people can see all my weaknesses	0	1	2	3	4
17. I feel like beating myself up when I make a mistake	0	1	2	3	4
18. Overall, I'm happy with who I am	0	1	2	3	4
19. When I make a mistake I wish I could just disappear	0	1	2	3	4
20. I go over bad things in my mind again and again until it gets too much.	0	1	2	3	4
21. I feel like I am a good person	0	1	2	3	4
22. At times, I feel I like I will fall apart	0	1	2	3	4
23. I feel that I have no control over my body and my feelings	0	1	2	3	4
24. Sometimes I feel no bigger than a pea	0	1	2	3	4
25. At times I feel like people can see right through me	0	1	2	3	4
26. I feel like I have a painful gap inside me that I'm not able to fill	0	1	2	3	4
27. I feel empty and unfulfilled	0	1	2	3	4
28. I have a positive attitude towards myself	0	1	2	3	4
29. My loneliness is more like emptiness	0	1	2	3	4
30. I feel like there is something missing	0	1	2	3	4

This is the end of the questionnaire

V13.7.9

Appendix T

Data Collection:
Patient Information Sheet for 8-11 Year Olds

Great Ormond Street Hospital for Children
NHS Trust

Great Ormond Street
London WC1N 3JH

Tel: 020 7405 9200

Information Sheet for Patients (8-11 years)

An exploration of psychological and social factors that contribute to adjustment in young people who look different.

What is this about?
We are trying to find out from children, what it's like to have a skin condition that makes you look different from other people. We know that some children who look different can get upset sometimes about it or sometimes find it more difficult to make friends. We want to ask as many children as possible what it's like and what helps to make it easier to live with.

What will you have to do?
All you have to do is fill out some questionnaires and do an interview the next time you come to Great Ormond Street Hospital for a check-up. You can do it while you wait to see the doctors and your parents will be with you to help you if you like. There will be an assistant who will help you with anything that you are not sure about. It should take you about 30-45 minutes to finish the questionnaires and about 30 minutes to do the interview. There are no right or wrong answers. It's just about how you feel about things. The interview will be video-taped so that we can remember everything that you say.

Your parents and school teacher will also be asked to fill out some questionnaires.

Why are we asking you?
We are asking all young people between 8 and 16 years of age who have a skin condition that makes them look different who come to Great Ormond Street Hospital.

Do I have to take part?
No. It is up to you and your parents to decide. If you decide you don't want to, that's fine. The doctors and nurses will look after you just the same as ever.

What about the results of the questionnaires and interview?
Your name will not be written onto any of the information that we keep about you. Once we get the information that we need from the video-tape of your interview, we will wipe the tape so that no-one else can see it. Therefore, no-one will know what answers you gave. All of your answers will be recorded onto our computer. Again, your name will not be stored with your answers. No-one will ever be able to find out what you told us.

Who will know about the results of the project?
When we finish, we will put all the answers together and try to work out how young people feel about looking different. This information will be given to you and your parents to look at. The results will also be shared with other doctors and nurses who

look after people like you because it might help them to be more aware of the way that young people feel about looking different. Most importantly, we hope that the information you and the other children and young people give us will help Great Ormond Street provide a better service to all people who come here.

Who can I speak to if I have any questions?
You can speak to your parents. They have been given information about this project. You can also speak to any of the doctors or nurses at Great Ormond Street. One of the people involved in this project is Kristina Soon, the clinical psychologist for Dermatology. You and your parents can always speak to her if you have any more questions.

Your parents have been given some other contact details of people to speak to if they have any questions or worries.

What happens now?
In about a week, the assistant will contact your parents by telephone to answer any questions you may have about this project and to ask if you would like to take part. If you agree, the assistant will explain what will happen next. You can change your mind, at any time, about whether you want to participate or not.

Appendix U

Data Collection:
Patient Information Sheet for 12-16 Year Olds

Great Ormond Street Hospital for Children
NHS Trust

Great Ormond Street
London WC1N 3JH

Tel: 020 7405 9200

Information Sheet for Patients (12-16 years)

An exploration of psychological and social factors that contribute to positive adjustment in young people who look different.

What is this about?
We know that it can be quite a challenge having a medical condition that makes you look different. We've already been collecting information about what it's like to look different and you may have already filled in some questionnaires as part of that. Now, we want to try to understand how family relationships and friendships can help people to cope with looking different. If we can work out what helps young people to cope well, we can use this information to help the young people who aren't coping quite so well.

What will you have to do?
All you have to do is fill out some questionnaires and do an interview the next time you come to Great Ormond Street Hospital for a check-up. You can do it while you wait to see the doctors and your parents will be with you to help you if you like. There will also be a researcher who will explain what you have to do and will be able to help you with any tricky questions. It should take you about 30 minutes to finish the questionnaires. Most of the questionnaires just ask you to tick the answer the best fits you and there are no right or wrong answers. The interview will also take about 30 minutes and you will be asked questions about how you deal with difficult situations. We will video-tape the interview so that we can remember everything that you say.

Your parents and your form teacher will also be asked to fill out some questionnaires.

Why are we asking you?
We are asking as many young people as possible between 8 and 16 years of age who have a medical condition that makes them look different and who come to Great Ormond Street Hospital for check ups and treatments.

Do you have to take part?
No. It is up to you and your parents to decide. If you decide you don't want to, that's absolutely fine. The doctors and nurses will look after you just the same as ever.

What about the results of the questionnaires and the interview?
Your name will not be written onto any of the questionnaires you fill out or onto the recording of your interview. Therefore, no-one will know what you said. Once we take the information that we need from the video-tape of your interview, we will wipe the tape so that no-one else can see it. All of your answers will be recorded onto our computer but your name will not be stored with your answers. No-one will ever be able to find out what you said.

Who will know about the results of the project?

When the project is finished, we will put all the answers together and try to work out how young people feel about looking different and what factors might help them to cope better. This information will be sent to you and your parents. The results will also be shared with other doctors and nurses because it might help them to be more aware about the feelings of young people who look different and how to support these young people best. Most importantly, we hope that the information you and the other young people give us will help Great Ormond Street provide a better service to all young people with medical conditions that make them look different.

Who can you speak to if you have any questions?
You can speak to your parents. They have been given information about this project. You can also speak to any of the doctors or nurses in Dermatology. One of the people involved in running this project is Kristina Soon, the clinical psychologist who works in Dermatology. You and your parents can always speak to her if you have any more questions.

Your parents have been given some other contact details of people to speak to if they have any complaints or worries.

What happens now?
In about a week, one of the researchers will contact your parents by telephone to answer any questions you may have about the project and to ask if you would like to be involved in this project. If you agree to participate, the researcher will explain what will happen next. You are free to change your mind, at any stage, about whether you want to take part or not.

Appendix V

Data Collection:
Parent Information Sheet

Great Ormond Street Hospital for Children NHS Trust

Great Ormond Street
London WC1N 3JH

Tel: 020 7405 9200

Information Sheet for Parents

An exploration of psychological and social factors that contribute to positive adjustment in young people who look different.

What is this about?
Great Ormond Street Hospital (GOS) is in the process of studying several factors that might help us to understand how young people cope with looking different. You and your child may already have been involved in the first stage of this project that looked at how young people with disfiguring medical conditions feel about their own appearance and their experience of how people react to them. We are now in the second phase of the project that focuses on the role of family and friends in helping young people to cope with looking different.

Why are we doing this?
There is a bit of research that shows that looking different can be difficult for young people to deal with, with many children and adults reporting problems, particularly, in social relationships. However, many young people cope very well and have excellent social relationships, despite looking different. We hope that if we can identify the factors that help young people to cope well, we can use this information to support the young people who are coping less well.

How will we do this?
We are asking young people who are have a dermatological condition that makes them look different, and their parents, to complete a small number of questionnaires the next time they come to GOS for an out-patient appointment**.** We would also like to do a video-taped interview with each young person, about how they deal with difficult social and family situations. The questionnaires will take about 30-40 minutes to complete and the interview about 30 minutes. We anticipate that questionnaires and interview can be completed while waiting for the appointment. One of the research team will be on hand to conduct the interview and to help out with any difficulties.

We would also like to send a few questionnaires to the school teacher of the young person. School is the main place that children and young people come into contact with other children and adults, and it would be helpful for us to get a clearer sense of how they are coping in that environment too.

Are there any risks involved?
As this is a non-invasive task; just questionnaires and an interview, there are very few potential dangers or risks to the participants. However, because some of the questions may seem quite personal or sensitive, such as questions about how they get on with other children, there is a possibility that some participants may feel upset or sad as they answer certain questions. If this happens, the researcher, who is a qualified psychologist, will be nearby to provide support. If more support is necessary any participant, child or parent, will be able to meet with the clinical psychologist attached to the dermatology Team at GOS for further assessment or support.

What about the results of the questionnaires and interviews?
Because we believe that this information is of a sensitive nature and should remain private, the name of the participant will not be attached to any of the information that we collect. The video-taped interviews will be transcribed into written form that will be anonymised and the video-tapes will be destroyed at the end of the study. Only the researchers will ever see the video-tapes of the interviews. Therefore, it will be impossible to provide individual results to participants and the results with remain confidential. Confidentiality will only be breached if a disclosure is made that indicates significant risk of harm to the individual or to someone else by the individual. However, this is a very rare occurrence and any break of confidentiality will be discussed with you and your child first.

The overall results of the study will be shared with all participants at the end of the project when the results have been analysed. The overall results will also be published in a scientific journal so that other professionals and organisations can benefit from the new information. Again, no information that is shared will reveal the identity of the individuals who participated in the study.

Who will have access to the completed questionnaires and interviews?
The completed questionnaires and interviews are "owned" by the GOS Psychosocial and Family Service and only people belonging to this team, who have been directly involved with this project, will be able to have a look at the information if they want to. Because names will not be attached to the questionnaires, no-one will know which participant completed each questionnaire.

The paper copies of the questionnaires, the recordings of the interviews and any data entered onto the computer system will be stored safely according to the Data Protection Act (1998). A representative of the hospital's Research Ethics Committee will also have access to data. If you have any questions about data protection, please contact the Data Protection officer via the switchboard on 020 7405 9200 Ext 5217.

Does my son or daughter have to take part in this project?
No. If you decide not to take part in this project, this is entirely your right and will not in any way affect your child's present or future treatment.

What are the arrangements for compensation?
This research project has been approved by an independent Research Ethics Committee that believes that it is of minimal risk to your child. However, all research can carry unforeseen risks and we want you to be informed of your rights in the unlikely event that any harm should occur as a result of taking part in this project.

No special compensation arrangements have been made for this project but you have the right to claim damages in a court of law. This will require you to prove a fault on the part of the hospital.

No participants will be offered payment of any kind or reimbursement of expenses in exchange for participating in this project as participation should not involve any additional costs to those you would normally incur coming to GOS for your appointment. All participants will be entered into a raffle, however, and could win a small cash prize for their involvement in the project.

Who do I speak to if I have further questions or worries?

Please contact Kristina Soon who is leading this project. You can contact her either through the GOS switchboard on 020 7405 9200 or by contacting the Department of Psychosocial and Family Services on 020 7829 8896.

If you have any complaints about the way in which the project is being or has been conducted, in the first instance please discuss them with Kristina. If the problems are not resolved, or you wish to comment in any other way, please contact Jo Southern, Head of Research and Development, Institute of Child Health, 30 Guilford Street, London WC1N 1EH or, if urgent, by telephone on 020 7242 9789.

What happens now?
In about a week, one of the research team will contact you by telephone to answer any questions you may have about the project and to ask you if you would like to take part. If you agree to participate, the researcher will explain what will happen next. You are free to change your mind, at any stage, about whether you want to participate or not.

If you do eventually participate in this project, your GP will receive a standard note informing them of your involvement. This note will not contain any personal information aside from your child's name, address and date of birth.

Version 3.9.3.9

Appendix W

Data Collection:
Letter of invitation to participate in the study

Great Ormond Street Hospital for Children
NHS Trust

Great Ormond Street
London WC1N 3JH

Tel: 020 7405 9200

"Date"
"Name and address of recipient"

To the Parents/ Guardians of

Re: Research Project for Young People who look Different

A project, run by Great Ormond Street Hospital (GOS) and University College London (UCL), has been set up to help us to understand what helps children to cope when their appearance is different as a result of a dermatological condition.

You may already have been involved in the first phase of this study, which looked at how young people with a dermatological disfigurement felt about themselves and how they looked. We are now going to look more closely at the role of family and friends on how these young people cope. For this study, we would like to get the opinions and experiences of as many young people as possible, between the ages of 8 and 16**.** As such, we would like very much for you and your son or daughter to take part.

We all know that looking different can be very challenging for anyone at any age. Surprisingly, there has been very little scientific research into this issue in young people so far. By finding out this kind of information, from as many young people as possible, we hope to be able to develop our psychology service here at GOS to meet the needs of our patients who look different, and their families, in a more effective and sensitive way. We also hope to be able to share this information with people working with similar children all around the world so that they can develop their services too.

There is are two Information Sheets attached to this letter that explain the details of the project; one for parents and one for the young person. They explain what we would want you and your child to do if you agreed to take part. Please read the Information Sheets carefully. One of the researchers will be in touch with you over the next few days to answer any questions you may have about the project in order to help you decide if you would like your son or daughter to participate. In the meantime, you are welcome to contact us on the telephone numbers below if you have any queries about the project.

Yours sincerely

Appendix X

Data Collection:
Consent form for parent or guardian

Great Ormond Street Hospital for Children NHS Trust and Institute of Child Health
Research Ethics Committee

Consent Form for PARENTS/GUARDIANS Participating in Research Studies

Title: An exploration of psychological factors that contribute to positive Adjustment in young people who look different.

NOTES FOR PARENTS/GUARDIANS

1. You have been asked to take part in a research project. The person organising the research must explain the project to you before you agree to take part.

2. Please ask the researcher any questions you like about this project, before you decide whether to join in.

3. If you decide, now or at any other time, that you do not wish to be involved in the research project, just tell us and we will stop the research. Your treatment will carry on as it would normally.

4. You will be given an information sheet which describes the research. This information is for you to keep and to read at any time. *Please read it carefully.*

5. If you have any complaints about the research project, discuss them with the researcher. If the problems are not resolved, or you wish to comment in any other way, please contact Jo Southern, Head of Research and Development, Institute of Child Health, 30 Guilford Street, London, WC1N 1EH or, if urgent, by telephone on 020 7242 9789.

CONSENT

I _____ agree that the Research Project named
above has been explained to me to my satisfaction, and I agree to take part in this study.
I have read both the notes written above and the Information Sheet about the project, and
understand what the research study involves.

SIGNED	**PRINTED**	**DATE**
---	---	---
SIGNED (Researcher)	**PRINTED**	**DATE**
---	---	---

Version 2.9.3.9

NOTES FOR THE RESEARCHER

. Use this assent form, in addition to the one for parents and guardians, if the child can understand what you wish to do.

. It is your responsibility to ensure that the parents/guardians and child understand what the research project involves, both theoretically and practically. **You must allow sufficient time to do this.** Make sure that the relatives or child can contact you if they have additional questions.

. A copy of this completed form must be placed in the patient's clinical records, and a copy must be kept by you with the research records.

. If there are any unforeseen ethical problems with this study you must inform the [a representative of the sponsor].

Appendix Y

Data Collection:
Assent Form for Child or Adolescent Participant

Great Ormond Street Hospital for Children NHS Trust and Institute of Child Health
Research Ethics Committee

Assent Form for YOUNG PEOPLE (8-16 years) Participating in Research Studies

Title: An exploration of psychological factors that contribute to positive adjustment in young people who look different.

NOTES FOR YOUNG PEOPLE

1. You have been asked to take part in a research project. The person organising the research must explain the project to you before you agree to take part.

2. Please ask the researcher any questions you like about this project, before you decide whether to join in.

3. If you decide, now or at any other time, that you do not wish to be involved in the research project, just tell us and we will stop the research. Your treatment will carry on as it would normally.

4. You will be given an information sheet which describes the research. This information is for you to keep and to read at any time. *Please read it carefully.*

5. If you have any complaints about the research project, discuss them with the researcher. If the problems are not resolved, or you wish to comment in any other way, please contact **Jo Southern**, Head of Research and Development, Institute of Child Health, 30 Guilford Street, London, WC1N 1EH or, if urgent, by telephone on 020 7242 9789.

ASSENT

I _____ agree that the Research Project named
above has been explained to me to my satisfaction, and I agree to take part in this study.
I have read both the notes written above and the Information Sheet about the project, and
understand what the research study involves.

SIGNED	**PRINTED**	**DATE**
---	---	---
SIGNED (Researcher)	**PRINTED**	**DATE**
---	---	---

NOTES FOR THE RESEARCHER

. Use this assent form, in addition to the one for parents and guardians, if the young person can understand what you wish to do.

. It is your responsibility to ensure that the parents/guardians and child understand what the research project involves, both theoretically and practically. **You must allow sufficient time to do this.** Make sure that the relatives or child can contact you if they have additional questions.

. A copy of this completed form must be placed in the patient's clinical records, and a copy must be kept by you with the research records.

. If there are any unforeseen ethical problems with this study you must inform the [a representative of the sponsor].

Appendix Z

Descriptions of the Most Common Dermatological Conditions in the Current Sample

Z.1 Dermatological Categories of the Study Sample

There is a very large number of dermatological conditions that can be present at birth or in the first year of life. The study sample reported 26 different dermatological conditions that were grouped into six main categories; Vascular Tumours, Atopic Dermatitis, Primary Bullous Disorders, Naevii, Overgrowth Syndromes and Mastocytoses. Each diagnostic category is briefly described below.

Z.1.1 Vascular Tumours.

Vascular anomalies in childhood are typically categorized into two groups; vascular tumours and vascular malformations. In the current study, the dermatological conditions included in the Vascular Tumours grouping were port-wine stains and infantile haemangiomas.

Vascular tumours are most commonly infantile haemangiomas, sometimes referred to as strawberry marks. Mixed or deep haemangiomas tend to be less circumscribed in boundary and have a bluish-purple colouration. 50% of these lesions are found on the head and neck area. Haemangiomas are relatively common with 1-2.5% of newborns are identified as having a haemangioma with the incidence increasing to approximately 4% in the first year of life.

Port-wine stains are superficial capillary malformations that are characterized by vivid red, clearly circumscribed lesions sometimes flat against the skin or with a nodular, plaque appearance. The incidence of portwine stains in the US is relatively high estimated to affect 0.1-2% of live births (Kanada, Merin, Munden, & Friedlander, 2012).

The majority of Vascular Tumours are localized and pose no immediate or long-term threat. However, ulceration can occur in 15% of haemangiomas causing pain,

bleeding and loss of mobility. Complications can also arise from the location of the anomaly. Peri-ocular haemangiomas cause opthalmological problems in most cases while haemangiomas located near the airways can cause significant, sometimes life-threatening breathing problems (Bruckner & Frieden, 2011). Figure Z.1 shows a facial port-wine stain with some thickening on the right side of the face.

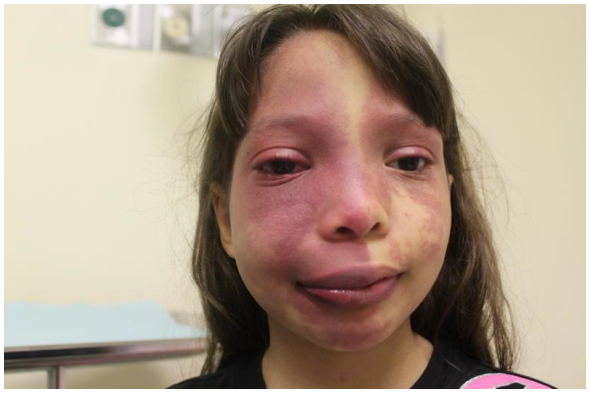


Figure Z.1 Girl with a Port-wine Stain with Hypertrophy (thickening of the skin) (Kids: Maricelli, n.d.)

Z.1.2 Atopic Dermatitis (AD) (Flohr & Williams, 2011)

AD often referred to as eczema, is typically defined as poorly demarcated erythema with surface changes such as scaling, vesicles, oozing, crusting or lichenification. It must be itchy and associated with involvement of the skin creases, history of asthma or hayfever (or history of atopy in a first-degree relative in the case of very young children), history of generally dry skin, onset before the age of two years and visible flexural dermatitis (Williams, Jburney, Strachan, Hay, Atopic Dermatitis Diagnostic Criteria Working, 1994)

Population surveys have varied in their estimation of prevalence due to differences in disease definition. Flohr & Williams (2011) report that morbidity figures for Western countries, including UK, range from 7-29%. Many of these cases will be self-managed or managed by general practitioners. A much smaller proportion of these children will attend specialist services (Schofield et al., 2009). Figure Z.2 shows typical lesions associated with Atopic Dermatitis.

Figure Z.2 Young woman with Atopic Dermatitis (Healing Atopic Dermatitis, n.d.)

Z.1.3 Primary Bullous Disorder

There are many environmental causes of skin blistering such as burns or excessive friction. Skin blisters can also be caused by acute disease such as Herpes Simplex or allergic contact dermatitis. Primary Bullous Disorder refers to a small number of genetic conditions that result in blisters from birth and across the lifespan in a recurrent pattern. The most common primary bullous disorder is Epidermolysis Bullosa (EB). Another bullous disorder is Bullous Icthyosiform Erythroderma (BIE). For the purposes of the current study, this diagnostic grouping includes participants with EB or BIE.

EB is a family of genetic disorders characterized by excessive susceptibility of the skin and mucosae to separate from the underlying tissue following mechanical trauma. Each form of EB varies in their impact from relatively minor and transient disability such as limitations to walking distance due to blistering of the feet to death in infancy (Mellerio & Denyer, 2011).

The most common and observable symptom of EB is blistering of the skin. Because EB can also affect the skin's capacity to heal, in many cases, people with EB can have open wounds for prolonged periods of time followed by skin scarring and joint contractures, which is permanent in some cases. EB can also cause nail and tooth

dysplasia. Because EB affects the mucosae, as well as the skin, blistering and scarring can be seen around the mouth, eyes, anus and genitals. Depending on the EB subtype, there can be multi-systemic complications such as including the gut, as well as pain and mobility problems as a result of the skin wounds.

It is estimated that there may be more than 5000 people in the UK with EB and possibly 500, 000 people worldwide (Debra UK, n.d.). Figure Z.3 shows siblings with EB Simplex.

Figure Z.3 Siblings with EB Simplex, a common, mild form of EB. (About EB, n.d.)

BIE is characterised by blisters, skin fragility and ichthyosis, which is the term that describes dry, thickened, scaly or flaky skin which is often characterized by deep cracking of the skin. The skin thickening can affect any part of the body but is most prominent on the scalp, around the neck and in the skin creases of the armpits, elbows and knees. Many patients with this condition develop thickening of the skin of the palms and soles. Older children and adults suffer from repeated skin infections especially in the skin folds (Icthyosis Support Group Uk, n.d.). BIE is a rare genetic skin disorder affecting fewer than 1 in 100,000 live births. Figure Z.4 shows typical skin thickening associated with BIE.

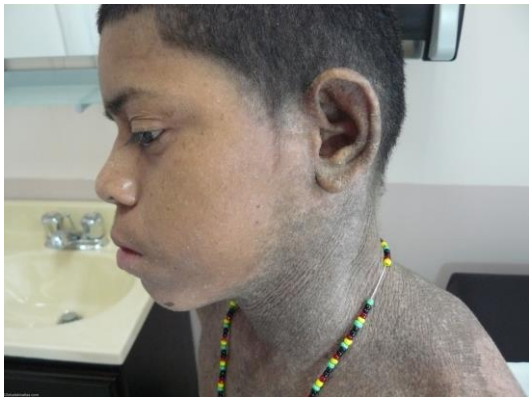


Figure Z.4 Young man displaying thickened, scaly skin characteristic of BIE (Epidermolytic Hyperkeratosis, n.d.)

Z.1.4 Overgrowth Syndromes

This diagnostic grouping encompasses a number of rare genetic disorders in which there is an abnormal increase in the size of the body or a single body part that is observable, usually, at birth. Types of overgrowth syndromes represented in the current study sample are neurofibromatosis, Proteus Syndrome and Klippel Trenaunay Syndrome.

Overgrowth Syndromes are characterized by overgrowth, asymmetry and gigantism of the limbs, organs or skeleton, raised rough skin or fat tissue. Complications to bodily functions and functional disability can occur depending on the location of the overgrowth. People who have overgrowth syndromes are also at significantly increased risk of blood clots and cancers that can be life threatening (www.proteus-syndrome.org.uk).

Overgrowth Syndromes tend to be extremely rare. For example, only 1000 people with Proteus Syndrome have been identified in the UK (Cohen, 2005). As such, this group of conditions are typically managed in specialist medical services. Figure X.5 shows a young woman with Neurofibromatosis Type 1.

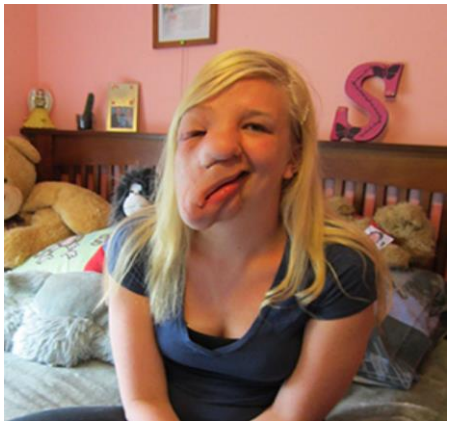


Figure Z.5 Young woman with Neurofibromatosis Type 1 (Lewiston Family Shares Story, n.d.)

Z.1.5 Naevii

Naevii are sharply circumscribed, dark lesions of the skin often referred to as beauty marks or moles. Many forms of nevus are acquired over the course of life such as through sun exposure.

Congenital naevii can be grouped into two categories; melanocytic naevii and epidermal naevii (Newton Bishop, 2011). Melanocytic naevii acquire their disclouration via the proliferation of melanocytes, the cells in the skin that cause pigmentation. Melanocytic naevii are often accompanied by proliferation of hair also. Congenital Melanocytic Naevii (CMN) are found in 1-2% of newborns (Osburn, Schosser, & Everett, 1987). Epidermal naevii, on the other hand, consist of keratinocytes or skin cells and are seen in two per 1000 live births. The majority of participants in the current study in the Naevii category suffer from CMN. Figure Z.6 shows a large CMN lesion on a young girl with typical proliferation of hair on the lesion.

Figure Z.6. Girl with Congenital Melanocytic Naevii (CMN). The largest lesion displays the characteristic proliferation of dark hair associated with CMN. (Congenital Melanocytic Naevii, n.d.)

Z.1.6 Cutaneous Mastocytosis

Mastocytoses encompass a range of manifestations caused by the presence of too many mast cells (*mastocytes*) and CD34+ mast cell precursors (Horny, Sotlar, & Valent, 2007). People affected by masyocytoses are susceptible to itching, urticaria, and anaphylaxis, caused by the excessive release of histamine from the mast cells. Cutaneous mastocytoses primarily involve the mast cells the skin and are typically characterized by patchy discolouration of the skin as well as the other symptoms caused by the presence of excessive mast cells (Léauté-Labrèze, Boralevi, & Taïeb, 2011). The mastocytoses represented in the sample of the current study were Urticaria Pigmentosa, Cutis Mamorata Telengactasia and diffuse Mastocytosis.

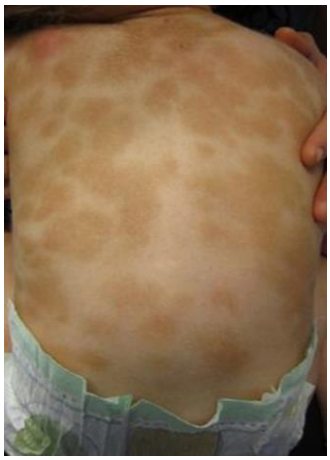


Figure Z.7 Back of child affected by Urticaria Pigmentosa (Urticaria Pigmentosa, n.d.)

Appendix AA

Within Group Differences: Attachment

Demographic Comparisons

Table AA.1
Gender

N= 99	Girls	Boys	X^2	df	*p*
Secure	33	21	.312	1	.576
Insecure	25	20			

Table AA.2
School Stage

N= 99	Primary	Secondary	X^2	df	*p*
Secure	25	27	1.792	2	.408
Insecure	17	29			

Table AA.3
Ethnicity

N= 96	White British	White Other	Asian	Black	X^2	df	*p*
Secure	48	4	0	1	4.975	3	.174
Insecure	38	1	3	1			

Table AA.4
Occupational Status

N= 95	Professional	Clerical & Intermediate	Senior Managers & Administrator	Technical & Craft	Semi-routine manual & craft	Routine manual & service	Middle & Junior Management	Unemployed	Value	df	*p*
Secure	18	1	14	6	3	0	6	5	12.586	8	.127
Insecure	15	2	10	5	2	3	4	1			

Comparisons By Medical Variables

Table AA.5
Disfigurement Location

N= 99	Face Only	Limbs Only	Torso Only	Whole Body	Face and Limbs	Limbs and Torso	Value	df	*p*
Secure	17	11	5	14	1	2	3.706	7	.813
Insecure	10	11	3	11	1	5			

Table AA.6
Dermatological Diagnosis

N= 99	Atopic Eczema	Vascular Birthmark	Bullous Disorders	Naevii	Overgrowth Syndromes	Masto-cytoses	Value	df	*p*
Secure	8	24	5	4	3	5	4.994	6	.551
Insecure	8	16	8	4	4	2			

Table AA.7
Age of Onset

N= 99	Birth	0-2 year	Value	df	*p*
Secure	44	10	.209	1	.648
Insecure	35	10			

Table AA.8
Hospital Attendance per Year

N= 93	< 1	1-2	3-5	6 or more	Value	df	*p*
Secure	3	36	8	5	3.411	3	.332
Insecure	1	23	12	5			

Table AA.9
Mobility

N= 96	Mobility Problems	Mobility Normal	Value	df	*p*
Secure	11	42	7.203	1	.007
Insecure	20	23			

Table AA.10
Pain

N= 97	Pain	No Pain	Value	df	*p*
Secure	13	40	.636	1	.425
Insecure	14	30			

Table AA.11
Medical Co-Morbidity

N= 97	Yes	No	Value	df	*p*
Secure	11	42	3.744	1	.053
Insecure	17	27			

Table AA.12
Life Threat

N= 95	Life Threat	No Threat	Don't Know	Value	df	*p*
Secure	8	43	1	1.664	2	.435
Insecure	10	31	2			

Appendix BB

Within Group Differences: Shame

Demographic Comparisons

Table BB.1
Gender

	Boys N=42	Girls N=69			
	X (SD)	X (SD)	t	df	*p*
External	33.06 (12.5)	30.51 (10.46)	-1.053	90	.371
Internal	22.78 (19.19)	19.42 (15.27)	-.848	53.843	.40

Table BB.2
School Stage

	Primary N=46	Secondary N=64			
	X (SD)	X (SD)	t	df	*p*
External	32.16 (9.86)	31.17 (12.13)	.395	89	.694
Internal	19.45 (12.83)	21.6 (18.99)	-.626	83.37	.533

Table BB.3
Ethnicity: Mean Scores

	White British N= 77	White Other N= 5	Asian N= 4	Black N= 1
	X (SD)	X (SD)	X (SD)	X (SD)
External	31.18 (11.12)	35.00 (14.71)	32.00 (16.02)	37.00 (.00)
Internal	19.92 (16.34)	25.00 (20.50)	25.25 (25.85)	40.00 (.00)

Table BB.4
Ethnicity: F Scores

	Sum of Squares	df	F	*p*
External	100.73	3, 85	.253	.859
Internal	593.38	3, 81	.683	.565

Table BB.5
OBBupational Status: Mean Scores

	Modern Professional	Clerical and Intermediate	Senior Managers & Administrator	Technical & Craft	Semi-routine manual and craft	Routine manual and service	Mid & Junior Management	Traditional Professional	Unemployed
	N=31	N=4	N=28	N=10	N=5	N=5	N=11	N=6	N=7
	X (SD)	X (SD)	X (SD)	X (SD)	X (SD)	X (SD)	X (SD)	X (SD)	X (SD)
External	29.26 (10.37)	46.67 (4.51)	32.35 (10.08)	29.67 (8.22)	30.75 (12.53)	28.75 (15.78)	37.75 (8.05)	25.33 (6.41)	35.71 (20.20)
Internal	18.86 (18.3)	25.25 (17.48)	21.57 (14.53)	15.71 (14.92)	14.75 (10.81)	27.00 (33.17)	29.25 (14.3)	15.5 (8.17)	25.17 (19.48)

Table BB.6
OBBupational Status: F Scores

	Sum of Squares	df	F	*p*
External	1542.19	8, 78	1.582	.144
Internal	1540.73	8, 74	.650	.733

Comparisons By Medical Variables

Table BB.7
Disfigurement Location: Mean Scores

	Face Only N= 29	Limbs Only N=20	Torso Only N= 7	Whole Body N= 19	Face+Limbs N= 2	Limbs+Torso N= 6
	X (SD)	X (SD)	X (SD)	X (SD)	X (SD)	X (SD)
External	29.45 (9.76)	30.60 (9.97)	28.71 (14.30)	35.45 (13.84)	25.00 (2.83)	34.33 (10.31)
Internal	17.21 (13.88)	19.75 (17.91)	20.71 (26.41)	20.68 (12.95)	16.50 (6.36)	22.83 (16.07)

Table BB.8
Disfigurement Location: F Scores

	Sum of Squares	df	F	*p*
External	943.72	7, 84	1.064	.394
Internal	3037.88	7, 79	1.618	.143

Table BB.9
Dermatological Diagnosis: Mean Scores

	Atopic Eczema N= 16	Vascular Birthmarks N=38	Bullous Disorders N=9	Naevii N=7	Overgrowth Syndromes N=6	Mastocytoses N=5	Other N=8
	X (SD)	X (SD)	X(SD)	X (SD)	X (SD)	X (SD)	X(SD)
External	33.87 (12.26)	28.79 (9.18)	35.30 (7.92)	34.83 (12.21)	29.00 (10.51)	34.67 (22.62)	33.57 (22.62)
Internal	25.44 (19.12)	16.42 (14.54)	23.38 (11.93)	19.00 (12.85)	22.33 (23.82)	19.75 (17.88)	29.13(22.12)

Table BB.10
Dermatological Diagnosis: F Scores

	Sum of Squares	df	F	*p*
External	732.17	6, 85	.956	.460
Internal	1719.62	6, 80	1.019	.419

Table BB.11
Age of Onset

	Birth N=71	0-1 N=19	t	df	*p*
	X (SD)	X (SD)			
External	31.39 (11.28)	31.83 (11.60)	-.148	90	.883
Internal	19.87 (16.70)	23.47 (17.24)	-.826	85	.411

Table BB.12
Hospital Attendance: Means Scores

	< 1 N=2	1-2 N=54	3-5 N=21	6 or more N=8
	X (SD)	X (SD)	X (SD)	X (SD)
External	38.50 (2.12)	32.23 (11.94)	27.95 (10.07)	33.57 (9.18)
Internal	20.50 (27.58)	21.74 (17.39)	13.33 (12.60)	26.88 (12.35)

Table BB.13
Hospital Attendance: F Scores

	Sum of Squares	df	F	*p*
External	422.32	3, 83	1.111	.350
Internal	1463.23	3, 77	1.895	.137

Table BB.14
Mobility

	Mobility Problems N=25	Mobility Normal N=63	t	df	*p*
	X (SD)	X (SD)			
External	34.88 (13.32)	30.22 (10.23)	1.792	88	.077
Internal	24.46 (17.35)	19.23 (16.66)	1.288	83	.201

Table BB.15
Pain

	Pain N=24	No Pain N=64	t	df	*p*
	X (SD)	X (SD)			
External	35.44 (9.20)	30.08 (11.78)	2.046	88	.044
Internal	26.70 (15.35)	18.48 (17.05)	2.025	83	.046

Table BB.16
Medical Co-Morbidity

	Yes N=28	No N=60	t	df	*p*
	X (SD)	X (SD)			
External	31.93 (9.93)	31.41 (11.96)	.196	88	.845
Internal	23.79 (17.50)	19.49 (16.57)	1.179	83	.242

Table BB.17
Life Threat

	Life Threat N=19	No Threat N=65	Don't Know N=2
	X (SD)	X (SD)	X (SD)
External	32.17 (11.38)	31.66 (11.59)	31.00 (5.66)
Internal	25.42 (20.67)	19.76 (15.82)	13.00 (5.66)

Table BB.18
Life Threat: F Scores

	Sum of Squares	df	F	*p*
External	4.78	2, 85	.018	.982
Internal	594.02	2, 80	1.034	.360

Appendix CC

Comparison Group Data Collection: Schools Information Sheet

Understanding the role of shame in children and adolescents: the relationship between shame and psychosocial functioning.

Background

The current study is part of a larger, ongoing research programme on children and adolescents with disfigurements; what their social development is like and how they cope psychologically with looking so different to everyone else. As part of this larger programme, we are keen to understand the role that shame plays in how young people develop socially and how they function psychologically. However, research into shame in children and adolescents is still in its early stages and we need to understand how shame functions for young people in the general population before we can properly understand how shame functions for young people who are disfigured.

The Study

Aim 1: Find out more about the levels and types of shame that children in the general population experience.

We expect that there will be a wide range of shame levels across different types of shame across all of the young people.

Aim 2: Find out if patterns of shame relate to patterns of social and psychological functioning.

We predict that shame will be correlated with social and psychological problems; high shame will be related to high psychosocial problems and low shame to low levels of psychosocial problems.

Data Collection

We would like to include as many children and adolescents as possible between the ages of 8 and 16 years.

The participants would be required to complete a set of questionnaires, which should take approximately 20 minutes.

We would have to exclude young people who are unable to complete the questionnaires, largely, on their own. This could be for reasons such as learning difficulties or English language difficulties.

One of the research team would liaise closely with the schools in the lead up to data collection in order to ensure that the process creates minimal disruption.

We would also hope to agree the consenting procedure with the school.

We hope to collect this data either before the summer holidays. We're happy to fit into the school's schedule to minimise disruption.

Results

All questionnaire information will be anonymous.
Once the data has been fully analysed, a short report can be sent to each participating school and the research team would be willing to explain the findings in more detail if helpful.

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